PSYCHOSEXUAL DEVELOPMENT, PSYCHOSOCIAL ADJUSTMENT AND QUALITY OF LIFE OF CHILDREN AND ADOLESCENTS WITH HYPOSPADIAS

Thesis presented to the Faculty of Arts of the University of Zurich
for the degree of Doctor of Philosophy

by

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Abstract

Objectives: To conduct a comprehensive cross-sectional study on health-related quality (HRQoL) of life, psychological adjustment and psychosexual development of children and adolescents after hypospadias repair and to identify sociodemographic, medical and psychosocial predictors of outcome. Prior to the actual investigation, a systematic review of previous research was conducted, which showed a lack of methodologically sound studies on the topic concerned.

Study design: Seventy-seven boys (7-17 years) operated on for hypospadias at the University Children's Hospital Zurich were investigated by means of a standardized interview. Patients mothers' were asked to fill out a questionnaire and medical data was retrieved from the patients' records. Scores were compared to an age-matched control group consisting of 77 boys after hernia repair. Predictive values of medical and psychosocial variables were assessed.

Main outcome measures: Child- and mother-rated HRQoL was evaluated by the *TNO-AZL Child Quality of Life Questionnaire* and psychological adjustment by the *Child Behavior Checklist.* The assessment instruments assessing psychosexual development comprised the self-developed *Pediatric Penile Perception Score*, the *Gender-role Questionnaire* by Ijntema and Cohen-Kettenis and a self-developed questionnaire on first sexual experiences and sexual attitude.

Results: Compared to the controls, self-reported HRQoL of hypospadias patients was lower in most dimensions. In contrast, mother-reported HRQoL and psychological adjustment did not differ between the two groups. Likewise, boys with hypospadias did not significantly differ from healthy controls with regard to penile self-perception, gender-role behavior, first sexual experiences and sexual attitude. Regarding predictors of outcome measures, higher age and "being less ashamed of penile appearance" predicted better self-reported HRQoL, while a positive penile self-perception, the patient's experience of not having been teased about his penis and more severe hypospadias contributed to better mother-reported HRQoL. Psychological adjustment was best predicted by higher age, Swiss nationality, more severe hypospadias and the patient's experience of not having been teased about his penis. With regard to psychosexual development, younger age and the patient's knowledge of hypospadias contributed to the prediction of a positive penile self-perception, whereas a more pronounced masculine gender-role behavior was best predicted by younger age at final surgery.

Conclusions: In contrast to psychological adjustment and psychosexual development, HRQoL of boys with hypospadias can be impaired. Overall, psychosocial factors such as the patient's age, his penile self-perception, his sense of shame, the reaction of peers and age-appropriate information about hypospadias may be more important predictors of the outcome than medical variables. However, the later corrective surgery is completed the more likely the patients may become insecure with regard to gender-role behavior.

Abbreviations

APA: American Psychological Association; CBAQ: Child Behavior and Attitude Questionnaire; CBCL: Child Behavior Checklist; CDI=Children's Depression Inventory; CGPQ=Child Game Participation Questionnaire; Cs=Cross-sectional; DPQ-J=Junior Dutch Personality Questionnaire; DSD: Disorders of Sex Development; EF: Encephalopathie Fragebogen; ES: Effect sizes; HRQoL: Health-related quality of life; ISNA: Intersex Society of North America; MIS: Mullerian inhibiting substance; MMPI=Minnesota Multiphasic Personality Inventory; Op.=Operation; PFK 9-14: Persönlichkeitsfragebogen für Kinder; PPPS: Pediatric Penile Perception Scale; PPPS Patient: patient's penile self-perception; PPPS Urologists: quality of surgical result; Proxy=Proxy-report measures; SAS-C=Social Anxiety Scale for Children; Self=Self-report measures; SES: Socioeconomic status; Sex.=Sexual; STAIC=State-Trait Anxiety Inventory; TACQOL-CF/-PF: TNO-AZL Child Quality of Life Questionnaire child/parent form; WMD: Weighted Mean Differences; YSR=Youth Self Report

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Tables and Figures

1 General Introduction

The aim of the present doctoral thesis was an extensive examination of psychological adjustment, health-related quality of life (HRQoL) and psychosexual development of children and adolescents with hypospadias - the most common penile malformation. Prior to a discussion of the currant state of research in chapter 2, the presentation of results in chapter 3 and 4, and an overall summary and final discussion of results in chapter 5, the present introductory chapter shall give some background information on the topic concerned. It includes an overview of the medical basics and assumed psychological consequences of hypospadias as well as provides the most important background information about this study, which was conducted at the University Children's Hospital Zurich. An initial explanation of the embryologic development of the male external genitalia is followed by a discussion of the classification, etiology and incidence of hypospadias. Furthermore, associated anomalies and the treatment of hypospadias will be discussed and various theses about the psychological effects of hypospadias shall be summarized. The provided background information on the present research project contains a detailed description of the objectives, hypotheses and applied methods of the study.

1.1 Embryologic Development of Male External Genitalia

During normal sexual differentiation, the chromosomal sex (♀: 46,XX; ♂: 46,XY) determines the gonadal sex (♀: ovaries; ♂: testes). The former is established at the moment of fertilization. Embryos which contain a XY chromosome are assumed to have a specific gene known SRY. This so-called testis-determining-factor (TDF) is situated on the distal end of the short arm of the Y chromosome. The TDF is believed to control the transcription of a series of genes necessary for the development of the testes from the undifferentiated gonads. The gonadal sex then determines the phenotypic sex: the appearance of the external genitalia (Sinnecker, 1999).

The formation of the external genitalia is a complex developmental process involving genetic programming, hormonal signaling, cell differentiation, tissue remodeling and enzyme activity (Baskin, 2004). The different stages of this process are outlined in figure 1. The early development of the external genitalia is indistinguishable in the two sexes. Up to the seventh week,

the external genitalia are undifferentiated structures constituting the ambisexual stage of sex differentiation (figure 1 A and B).
Although distinguishing sexual characteristics begin to appear during the ninth week, the external genital structures are not fully differentiated until the twelfth week of pregnancy (Moore & Persaud, 2007; Sinnecker, 1999; Yamada, 2003).

Early in the fourth week, proliferating mesenchyme (embryonic connective tissue) build a genital tubercle in both sexes at the cranial end of the cloacal membrane, the premordium of the penis in males and of the clitoris in females (figure 1 A). Labioscrotal swellings (genital swellings) and urogenital folds (urethral folds) develop shortly thereafter on each side of the cloacal membrane. These mesodermal components retain the capacity to convert testosterone to dehydrotestosterone. The genital tubercle soon elongates to form a phallus, which is as large in female as in male embryos (Duckett, 1998; Moore & Persaud, 2007; Seseke & Ringert, 2001).

As development proceeds, a wedge of mesenchyme, known as the urorectal septum, migrates towards the caudal end of the hindgut. When it fuses with the cloacal membrane at the end of the sixth week, it divides the cloacal membrane into dorsal and ventral parts (figure 1 B). While the dorsal portion forms the anal membrane, the ventral portion forms the urogenital membrane. The urogenital membrane lies in the floor of the urogenital groove, a median cleft, and is surrounded by the folds described earlier. Shortly thereafter, these membranes ruptures, forming the anus and urogenital orifice, respectively. The urethral groove begins to extend along the ventral surface of the phallus (Moore & Persaud, 2007; Seseke & Ringert, 2001; Westenfelder, 1993).

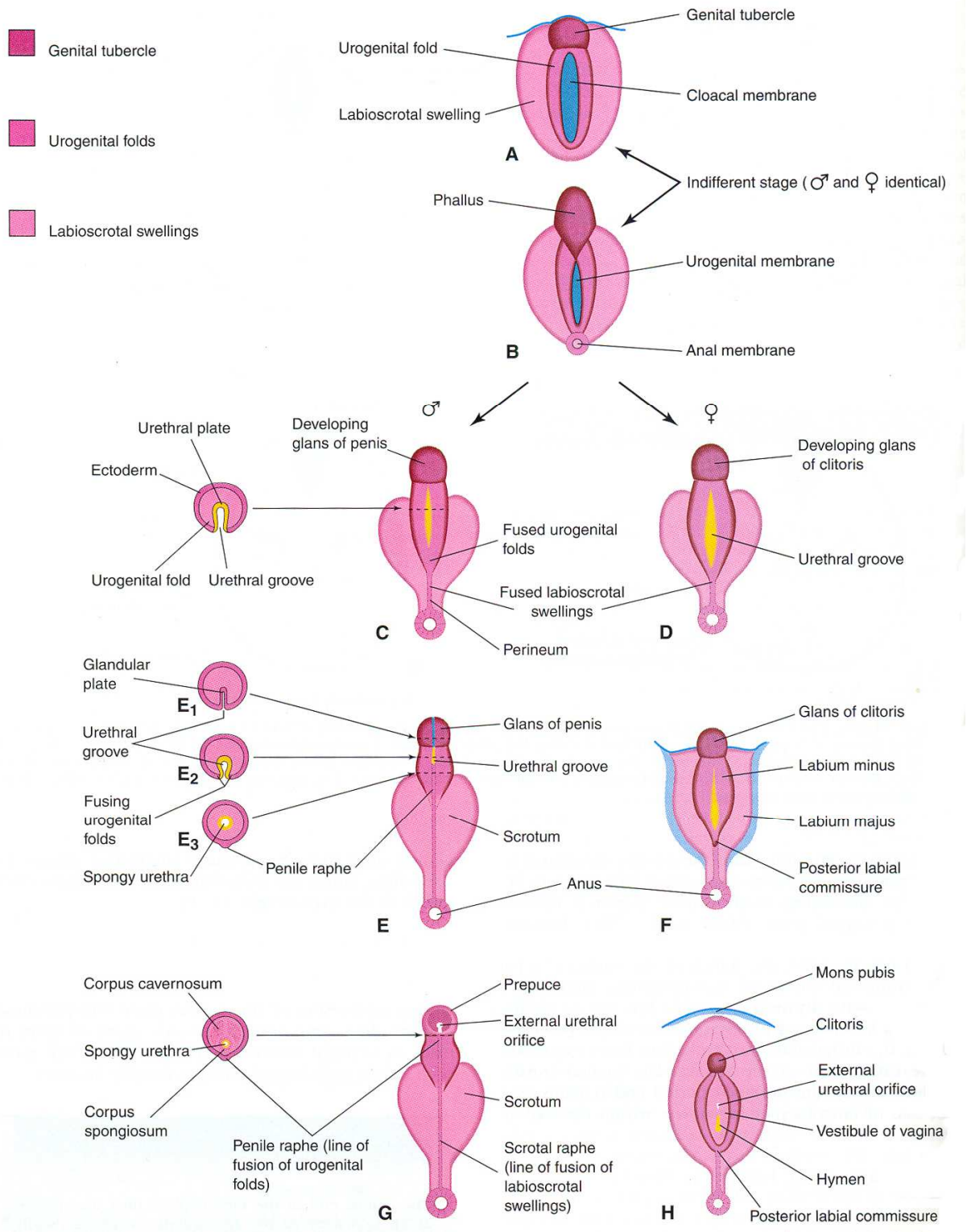

Figure 1: Development of the external genitalia. A and B, Diagrams illustrating the appearance of the genitalia during the indifferent stage (fourth to seventh weeks). C. E, and G, Stages in the development of male external genitalia at 9, 11 and 12 weeks, respectively. To the left are schematic transverse sections of the developing penis, illustrating formation of the spongy urethra. D. F, and H, Stages in the development of female external genitalia at 9, 11, and 12 weeks, respectively (Moore & Persaud, 1998, p. 332).

Around the eighth week of pregnancy, exposure to androgens begins (Moore & Persaud, 2007). The latter are produced by the fetal testes, which begin to develop at about the sixth week of gestation (Carillo, Damina, & Berkovitz, 2007). Under the influence of androgens, masculinization of the external genitalia takes part, which means that the latter develop a typical male appearance (Baskin, 2004; Duckett, 1998; Moore & Persaud, 2007). While the genital tubercle elongates and enlarges to form the penis, the urogenital folds begin to grow towards each other and form the lateral walls of the urethral groove on the ventral surface of the penis (figure1 A and B). This groove is lined by a proliferation of endodermal cells called the urethral plate (figure 1 C) (Moore & Persaud, 2007). The urogenital folds soon begin to fuse with each other along the ventral surface of the developing penis, enclosing what will now become the spongy urethra (figure 1 E1, E2, E3). The ectoderm fuses in the median plane of the penis, forming the penile raphe and enclosing the endoderm-lined urethra within the penis. As a consequence of these changes, the external urethral orifice migrates progressively towards the glans penis (Moore & Persaud, 2007; Seseke & Ringert, 2001; Westenfelder, 1993; Yamada, 2003).
Meanwhile, at the tip of the glans, a cord of ectoderm is developing called the glandular plate. The glandular plate grows towards the spongy urethra. When it becomes canalized and meets the previously formed spongy urethra, the terminal part of the urethra is completed and moves the external urethral orifice to the tip of the glans (Moore & Persaud, 2007).

The future foreskin is forming at the same time as the urethra and is dependent on normal urethral development (Baskin, 2004). A septum of ectoderm moves inward around the edges of the glans. When this cellular ingrowth breaks down or splits, it forms the prepuce, a thin layer of skin that surrounds the penis but is separated from the glans. However, for some time, the prepuce is adherent to the glans and is usually not retractable at birth. But subsequent breakdown of the adherent surface during infancy allows the prepuce to retract (Baskin, 2004; Moore & Persaud, 2007; Westenfelder, 1993). The corpora cavernosa penis and the corpus spongiosum penis arise from the mesenchymal tissue in the phallus. Approximately at the same time, the labioscrotal swellings grow towards each other and eventually fuse to become the scrotum. The line of fusion of the labioscrotal folds is clearly visible and is called the scrotal raphe (figure 1 G) (Moore & Persaud, 2007).

1.2 Hypospadias: Classification, Etiology and Incidence

Since the embryologic development of the male external genitalia described in chapter 1.1 is a complex process, developmental failures can occur at each of the different stages of external sex determination. Such failures can result in various genital anomalies. Thereby, the most common malformation of the penis is hypospadias, which is manifested by an abnormal position of the urethral opening. The latter lies proximal to its normal location in the glans, on the ventral surface of the penis or in the perineum (figure 2).

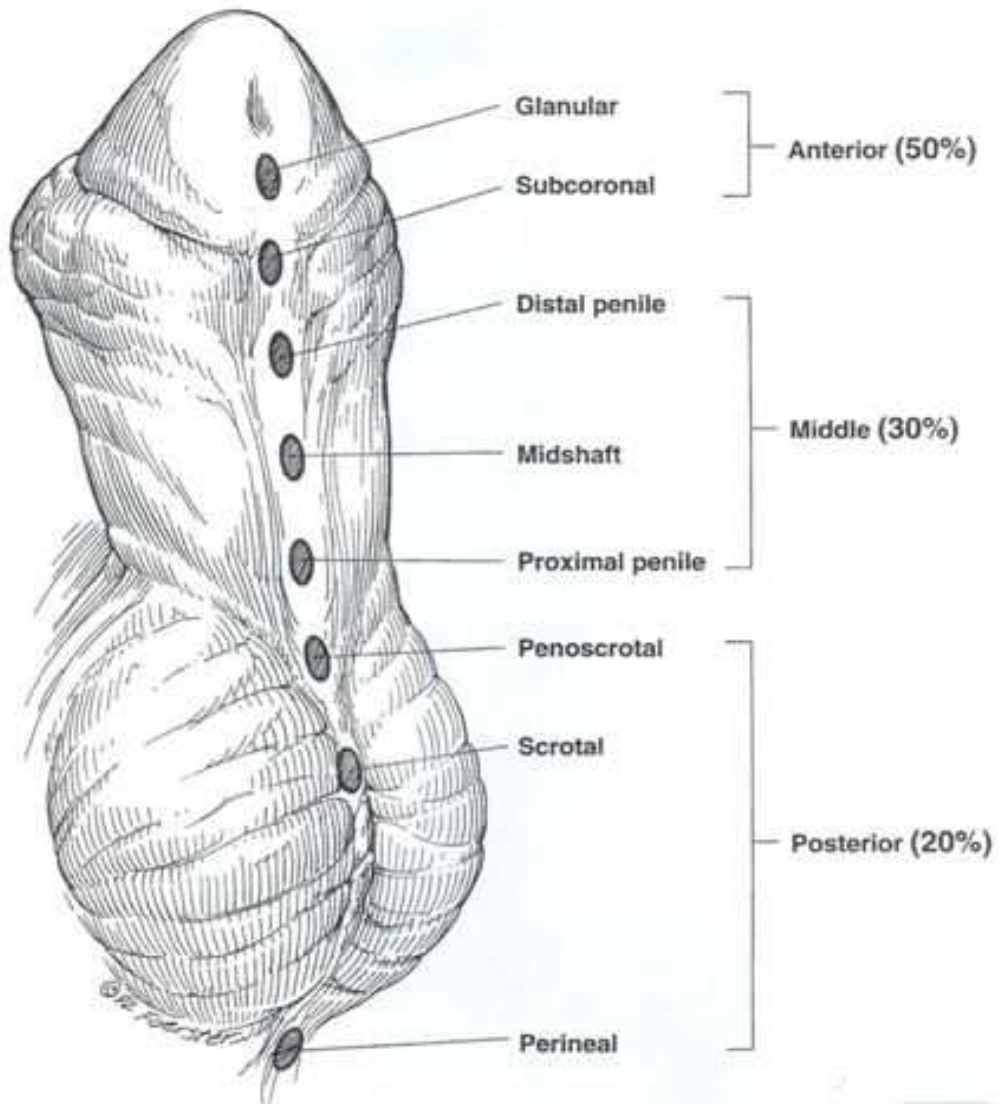

Figure 2: Classification of hypospadias according to Duckett (1998, p. 2094)

A prepuce that is split on the ventral side, a curvature of the penis (chordee) and a bifid (when the two halves of the scrotum meet above the penis) or transposed scrotum are associated findings in hypospadias (Duckett, 1998; Manzoni, Bracka, Palminteri, & Marrocco, 2004). In the commonly used classification system of hypospadias, the position of the meatus classifies the severity of hypospadias: The more proximal the meatus, the more severe the hypospadias. This common method of classification of hypospadias is currently criticized since it does not involve the degree of penile curvature. It is therefore recommended to classify hypospadias not on the basis of the original site of the meatus but on the basis of its new location after surgical correction of chordee (Duckett, 1998; Hadidi, 2004). Figure 3 shows the penis of an infant with uncorrected proximal penile hypospadias and associated penile curvature.

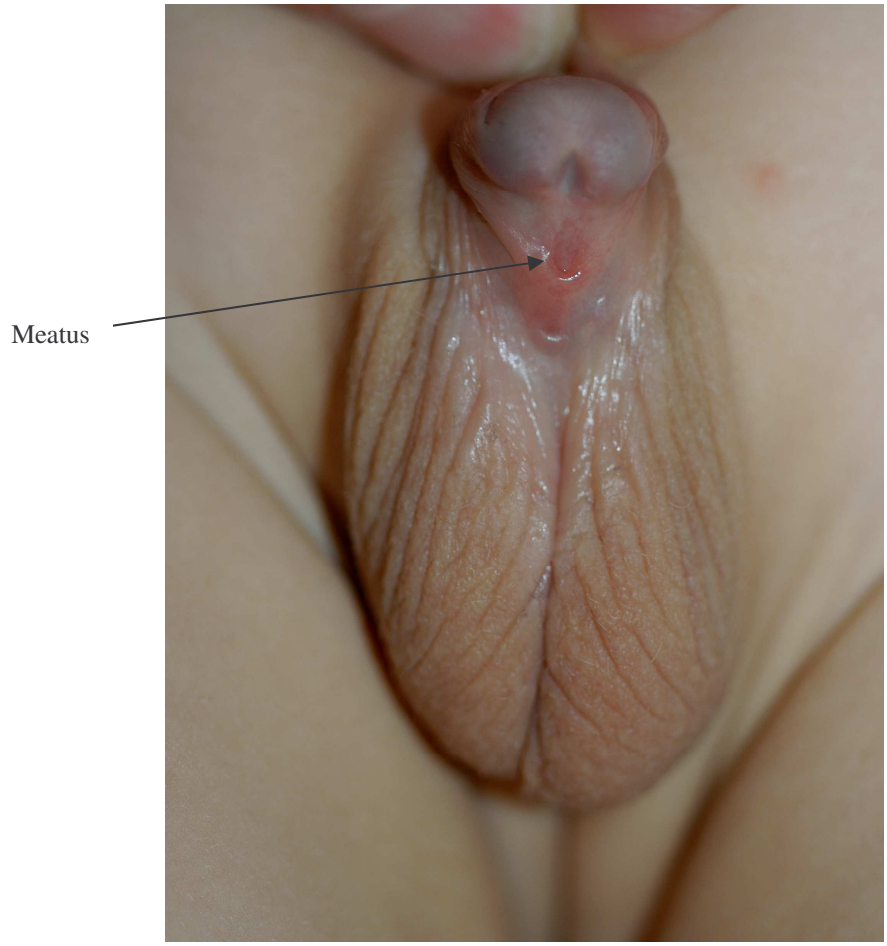

Figure 3: Penis of an infant with uncorrected proximal penile hypospadias and associated chordee

The underlying etiological factors of hypospadias are not exactly known. So far, most explanations have focused on genetic, endocrinological and environmental factors. However, although the etiology of hypospadias is still hardly intelligible, it is still clearly evident that hypospadias is the result of a faulty embryogenesis of the penile urethra related to abnormalities in prenatal androgen synthesis and/or androgen receptor defects (Baskin, 2004; Duckett, 1998; Moore & Persaud, 2007). These defects lead to a failure of canalization of the glandular plate and/or failure of fusion of the urogenital folds on the ventral side of the penis. As a consequence, the formation of the urethral groove into a tubular penile urethra is incomplete and the urethra does not reach the top of the penis (Moore & Persaud, 2007; Yamada, 2003). Differences in the timing and degree of hormonal malfunctioning determine the precise site of failure of the formation of urethral fold fusion and thus will dictate the location of the abnormal opening of the urethra: from perineal positions proximally to more distal openings along the shaft of the penis. (Moore & Persaud, 2007; Seseke & Ringert, 2001; Westenfelder, 2003).

The malformation of the foreskin in hypospadias is related to the abnormal urethral development. In hypospadias the normal scrotal and penile raphe divides into a Y shape running onto the dorsal preputial hood around the glans in varying configurations. Thus, failure of the urethra to reach the tip of the glans is accompanied by the absence of the ventral foreskin (Carillo et al., 2007; Duckett, 1998). The most poorly understood entity in hypospadias, however, is the penile chordee (Duckett, 1998). According to Duckett, it is most likely that penile curvature is the result of a growth differential between the normally formed dorsal tissue of the corporal bodies and the deficient ventral tissue as well as the underdeveloped urethra.

Reports on the incidence of hypospadias are inconsistent and vary considerably across different countries (Dolk, 2004; Paulozzi, 1999). Dolk (2004) estimates a maximal incidence of approximately 3 per 1000 male births in western countries. There is also some evidence that the prevalence of hypospadias had been increasing during the 1960s, 1970s, and 1980s (Dolk, 2004). Recently, Paulozzi (1999) analyzed data from the International Clearinghouse for Birth Defects Monitoring System (ICBDMS), a non-governmental organization of the World Health Organization (WHO). Aim of the study was to examine whether the observed increases in the incidence of hypospadias were worldwide continuing and whether they revealed any geographic pattern. The results indicate that increases were most marked in the United States, Scandinavia and Japan but were not seen in less industrialized nations.

The increased occurrence of hypospadias has raised questions with reference to the etiology of hypospadias. It has been hypothesized that the observed increase might have been caused by exposure to endocrine-disrupting chemicals (Dolk, 2004; Paulozzi, 1999). The latter are exogenous substances that interfere with the endocrine system and consequently lead to adverse effects on health. Such substances occur not only in the occupational setting but also in the general environment, in food packaging and in food itself (Dolk, 2004). With regard to hypospadias, the healthy development of the penile urethra, which is dependent on a sufficient androgen synthesis, is speculated to be impeded by anti-androgenic chemicals (Dolk, 2004; Paulozzi, 1999). However, it is also assumed that some of the changes in the incidence of hypospadias might be artefacts of study methodology. For instance, it is thought that the rising occurrence reflects the gradually changing definition of hypospadias to include more minor degrees of hypospadias. However, reports on rising rates of hypospadias correspond with reports on increasing incidences of potential aetiologically related conditions such as cryptorchidism and testicular cancer. These observations support the hypothesis that environmental factors may play a role in the etiology of hypospadias (Baskin, 2004).

1.3 Associated Anomalies

Hypospadias is often associated with further anomalies. The most common associated anomalies are undescended testes and inguinal hernia. Incidence of both anomalies is about nine percent in patients with hypospadias and is higher in more severe forms of hypospadias than in mild forms (Baskin, 2004; Duckett, 1998). A utriculus masculinus, a derivative of the mullerian ducts, is sometimes detected in severe forms of hypospadias (Baskin, 2004). The mullerian duct takes up an important part in the development of the female reproductive system. In normal male development, the mullerian ducts disappear under the influence of the mullerian inhibiting substance (MIS) (Moore & Persaud, 2007). Patients with hypospadias associated with utriculus masculinus may thus have insufficient secretion of MIS during intrauterine period (Kulkarni, Oak, Patel, Merchant, & Borwankar, 1991).

In contrast, significant urinary tract anomalies are infrequently associated with isolated hypospadias. This is unsurprising as the external genitalia are formed later during the embryologic development than the supravesical portion of the urinary tract (Baskin, 2004; Duckett, 1998). However, it is important to note that in more severe forms of hypospadias, the diagnosis of a disorder of sex development (DSD; recently introduced umbrella term for intersex states)

should be excluded (e.g. partial androgen insensitivity syndrome), especially when cryptorchidism coexists (Duckett, 1998).

1.4 Treatment of Hypospadias

The common treatment of hypospadias enfolds corrective surgery with the objective of achieving a normal looking penis, allowing micturition in the standing position and enabling normal sexual functions (Duckett, 1998). In mild hypospadias, however, cosmetic aspects (normal penile appearance) are often the only real indications for treatment (Manzoni et al., 2004).

Hypospadias surgery usually includes the reconstruction of the urethra, the straightening of the penis, the creation of a new meatus, skin coverage and the correction of any penile scrotal transposition and/or bifid scrotum (Baskin, 2004; Duckett, 1998). During the past decades, much research has been conducted to improve the surgical techniques. Meanwhile, over 200 operation techniques have been described and pediatric urologists have been constantly trying to optimize the surgical outcome. According to distinguished pediatric surgeons, hypospadias surgery has turned into a safe, reliable and highly successful procedure (Duckett, 1998; Manzoni et al., 2004). However, hypospadias repair still can produce unsatisfactory results, such as scarring, urethral fistulas (unwanted orifice through the skin alongside the urethra which interferes with urinary flow and may result in post-urination leakage), residual chordee (curvature of the penis), meatal stenosis (narrowing of the urethra which constricts urinary flow) and balanitis xerotica obliterans (chronic inflammation). Such complications tend to occur more often in severe than in mild hypospadias and predominantly require further surgical treatment (Duckett, 1998). Figure 4 shows the penis of an infant after successful hypospadias repair, figure 5 the penis of an infant with a postoperative fistula.

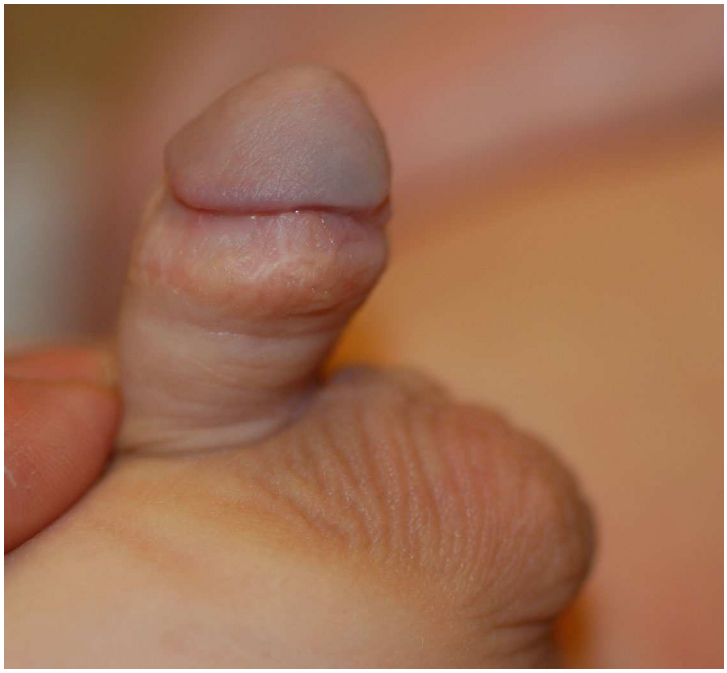

Figure 4: Penis of an infant after successful hypospadias repair

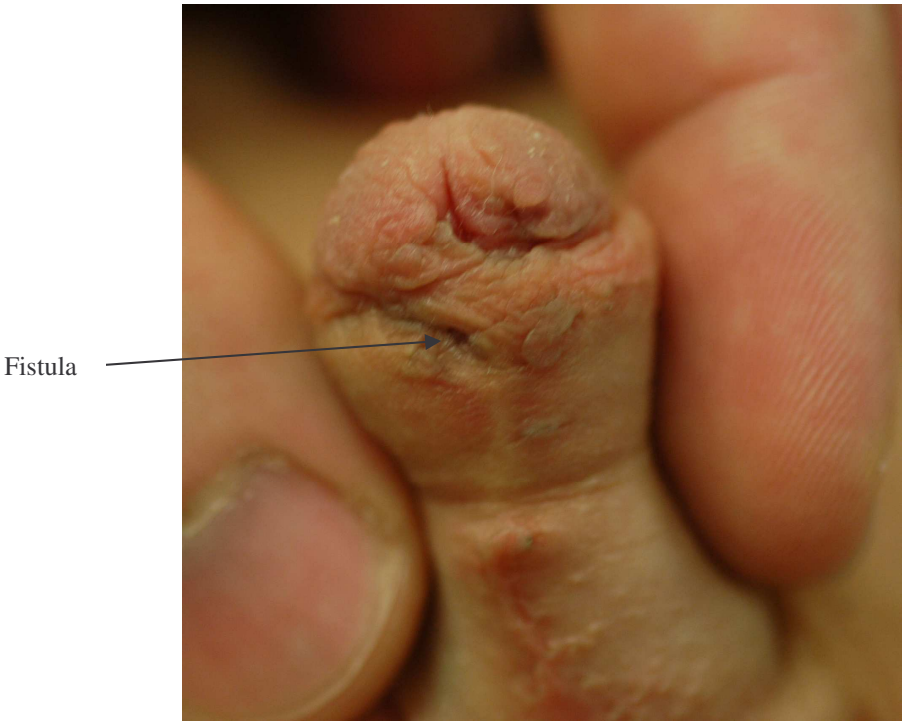

Figure 5: Penis of an infant with a postoperative fistula

1.5 Psychological Effects of Hypospadias

Several factors related to hypospadias are assumed to expose the patients to an increased risk of psychosocial and psychosexual maladjustment. Firstly, the patients generally experience the stressors of genital surgery and hospitalization, sometimes on multiple occasions (Mureau, Slijper, Slob, & Verhulst, 1997; Purschke & Standke, 1993; Sandberg et al., 2001). Secondly, hypospadias surgery never results in a perfectly normal penile appearance. Thus, the boys grow up with the awareness that their penis looks differently compared to others, which might affect their overall body image and ultimately lead to sexual withdrawal. Peer teasing due to atypical penile appearance may additionally enforce the boys' sexual inhibitions and sense of shame (American Academy of Pediatrics, 1996; Manzoni et al., 2004; Mureau, Slijper, Slob, & Verhulst, 1995). Likewise, as described above, hypospadias surgery can entail some functional complications, which mainly refer to urinating problems (Duckett, 1998). It is supposed that the ability to urinate in a typical male manner is essential for the boys to develop sufficient sexual self-confidence and to be socially accepted by their peers (Meyer-Bahlburg, 1999; Mureau et al., 1995; Sandberg et al., 2001; Zavitsanakis & Gougoudi, 2004). Thirdly, it is presumable that parents of children with hypospadias are worried about their son's future masculine capabilities. Such anxieties might result in an overprotection of the patient which in turn may limit his development of autonomy and self-confidence. Moreover, they might be transferred on the patient and consequently reinforce his negative appraisal of his penile appearance (Berg, Berg, & Svensson, 1982; Mureau et al., 1995). Finally, there is increasing evidence that prenatal exposure to androgens does not only induce the sexual differentiation of the human genitalia but also influences postnatal sex-typed behavior patterns by engendering sex-specific brain structures. Most of the evidence derives from studies on people with DSD, which are exposed to an atypical hormone synthesis (Cohen-Bendahan, van de Beek, & Berenbaum, 2005). Jürgensen, Hiort, Holterhus, and Thyen (2007) for instance have recently found a connection between prenatal hypoandrogenization and gender-related behavior in 33 children with XY Karotypes and DSD. As the etiology of hypospadias is related to abnormalities in prenatal androgen synthesis and/or androgen receptor defects, it has been hypothesized that children born with hypospadias, like children with some forms of DSD, might tend to atypical gender-role behavior (Sandberg et al., 1995). However, this hypothesis has not yet been confirmed.

A further indicator that patients with hypospadias experience specific psychological stress is the recently started formation of self-help groups of adult patients (e.g. Hypospadias and Epispadias Association, New York). The participation in self-help groups allow patients to relate to others with genital malformations and is thus believed to decrease the patients' senses of isolation and shame (Lee, Houk, Ahmed, & Hughes, 2006). A further objective of the groups is the support of the patients' needs' with regard to medical treatment. Recently, the majority of self-help groups have asked to reduce the number of surgical procedures for cosmetic reasons due to the potential of surgical complications to cause emotional trauma. The groups also emphasize the significance of age appropriate information of patients about their malformation (Intersex Society of North America, 1994, 2006; M. Walker, personal communication, December 13, 2006).

However, although marked psychological implications for children born with hypospadias seem to prevail, it should be also considered that surgical treatment strategies have evolved over the last decades: Surgical techniques and anesthesia have been improved and nursing practice has adapted to children's needs as for example by the rooming-in of parents (Bracka, 1999). These improvements are presumed to have diminished the potential risk of impaired psychosocial and psychosexual adjustment of patients with hypospadias. According to Bracka (1999), modern management of hypospadias is considerably less stressful than it used to be: "There is less physical trauma, with fewer complications, and better postoperative function and cosmesis. Hospitalizations are fewer and shorter, …. We anticipate that patients will grow up to have fewer psychological problems and negative feelings than their predecessors." (p. 30).

Furthermore, refined techniques and progress in pediatric anesthesia nowadays allow surgical repair in the latter half of the first year of life. It is believed that hypospadias should be corrected as early as possible because the penile malformation is assumed to impair the child's developing body image as well as his gender identity. Since it is thought on the one hand that the patient's awareness of both the different sexes and penile deformity occurs at about the 30th month of age, and on the other hand that surgical intervention is most likely to cause emotional trauma in children between the ages of one and three, hypospadias repair is recommended to be conducted between the sixth and twelfth months of life (American Academy of Pediatrics, 1996; Manzoni et al., 2004; Zavitsanakis & Gougoudi, 2004). It is, however,

important to note that these suggestions on the timing of surgical correction are not evidence-based.

The lack of empirical knowledge about the psychological significance of the patient's age at corrective surgery results from the fact that hardly any research has been conducted on the psychological and psychosexual development of children and adolescents with hypospadias. Most previous studies focused on the psychological and sexual consequences of hypospadias in adulthood (e. g. Aho, Tammela, Somppi, & Tammela, 2000; Berg et al., 1982). Moreover, many studies on both adult patients and children show methodological weaknesses (e. g. Kumar & Harris, 1994; Lepore & Kesler, 1978; Schubert, Kelly, & Trinckauf, 1989), such as absence of comparison data, small sample size and use of non-validated measures. Although more previous studies are methodologically higher qualified (Mureau et al., 1997; Sandberg et al., 2001), the overall low methodological standard of many studies might have contributed to the rather inconsistent findings concerning psychological adjustment and psychosexual development of children with hypospadias. A further limitation of most previous studies refers to the examination of potential risk factors. Although several studies have examined the influence of medical characteristics (e.g. severity of hypospadias) on the outcome, the association with psychosocial factors (e.g. parental attitude towards hypospadias) was neglected. A comprehensive systematic review of previous studies on the psychological well-being and psychosexual development of boys with hypospadias is presented in chapter 2.

However, regardless of the studies' methodological quality, inconsistent findings and neglected research questions, results of previous studies are considered to be obsolet due to the essential improvements in surgical and nursing treatment during the past few years (Mureau et al., 1997). As mentioned above, these improvements are supposed to have diminished the psychological consequences of hypospadias. Comprehensive examinations of the psychological adjustment and psychosexual development of boys with hypospadias are thus urgently required.

1.6 Project at the Children's University Hospital Zurich

The present Ph.D-project is based on a study on the psychological adjustment, health-related quality of life and psychosexual development of children and adolescents with hypospadias which was conducted at the Children's University Hospital Zurich between November 2005 and April 2007. The study was initiated by Dr. Daniel Weber (Department of Pediatric Urology), PD Dr. Markus Landolt (Department of Psychosomatics and Psychiatry) and PD Dr.

Rita Gobet (Department of Pediatric Urology) due to the lack of empirical knowledge on the psychosocial consequences of hypospadias in childhood and adolescence. The author of the present doctoral thesis was employed as a research fellow to conduct the study under the supervision of Dr. Daniel Weber and PD Dr. Markus Landolt. She made crucial contributions to the study design and was responsible for the development of the children's interview and mothers' questionnaire, for the recruitment of the patients, for the acquisition of data, for the statistical analyses and for the writing of three journal articles which mainly constitute the present doctoral thesis. The study at the Children's University Hospital Zurich was financially supported by the Foundation Mercator, Switzerland.

1.6.1 Objectives, Research Questions and Hypotheses

Aim of the study was an extensive examination of the psychological adjustment, health-related quality of life and psychosexual development of children and adolescents with hypospadias as well as of medical and psychosocial risk factors. Particular attention was paid to the examination of the patients' health-related quality of life (HRQoL) since the present study was the first to investigate HRQoL of boys with hypospadias. While psychological adjustment incorporates a range of outcome measures including behavioral and psychosocial factors (Achenbach, 1991), the multidimensional concept of HRQoL is defined as the patient's subjective evaluation of his/her physical, cognitive, social and emotional functioning. The concept is thus not solely confined to physiological outcome and/or psychopathological symptoms. Therefore, HRQoL has recently become clearly established as one of the most important approaches in assessing significant consequences of the patients' state of health (Edwards, Patrick, & Topolski, 2003; Matza, Swensen, Flood, Secnik, & Leidy, 2004).

In contrast, the concept of psychosexual development comprises many components such as the development of gender identity, gender-role behavior, body image, body inquiry, the development of sexual feelings, first sexual experiences and sexual behavior habits (Schuhrke, 1997; von Sydow, 1993). With reference to previous results and theoretical assumptions, we considered the following aspects of psychosexual development to be most important for our study: Penile self-perception, gender-role behavior, first sexual experiences and overall sexual attitude.

The study aimed at the examination of the following research questions and hypotheses:

1. Do children and adolescents with surgically corrected hypospadias show an impaired psychological adjustment, HRQoL and psychosexual development compared to healthy children and adolescents?

   H1: Children and adolescents with surgically corrected hypospadias show an impaired psychological adjustment, HRQoL and psychosexual development compared to healthy children and adolescents.

2. Are there any important medical, sociodemographic or psychosocial determinants of the outcome, such as the severity of hypospadias, age at corrective surgery, number of operations, number of days in hospital, follow-up since last surgery, quality of surgical result, current age, socioeconomic status, nationality, the patient's knowledge of hypospadias, his sense of shame and his experience of having been teased about his penis?

   H2: a) More severe hypospadias, higher age at corrective surgery, a higher number of operations, a higher number of days in hospital, a shorter follow-up since last surgery and a better quality of surgical result are associated with a more positive outcome, b) current age is negatively, socioeconomic status and Swiss nationality are positively associated with the outcome and c) patients' who are informed about their hypospadias, who are not ashamed of their hypospadias and who have not been teased about their hypospadias show a better outcome.

Figure 6 schematically depicts the most pivotal research questions and hypotheses of the study. It displays the influence of surgically corrected hypospadias as well as of medical, sociodemographic and psychosocial factors on the patients' psychological adjustment, HRQoL and psychosexual development.

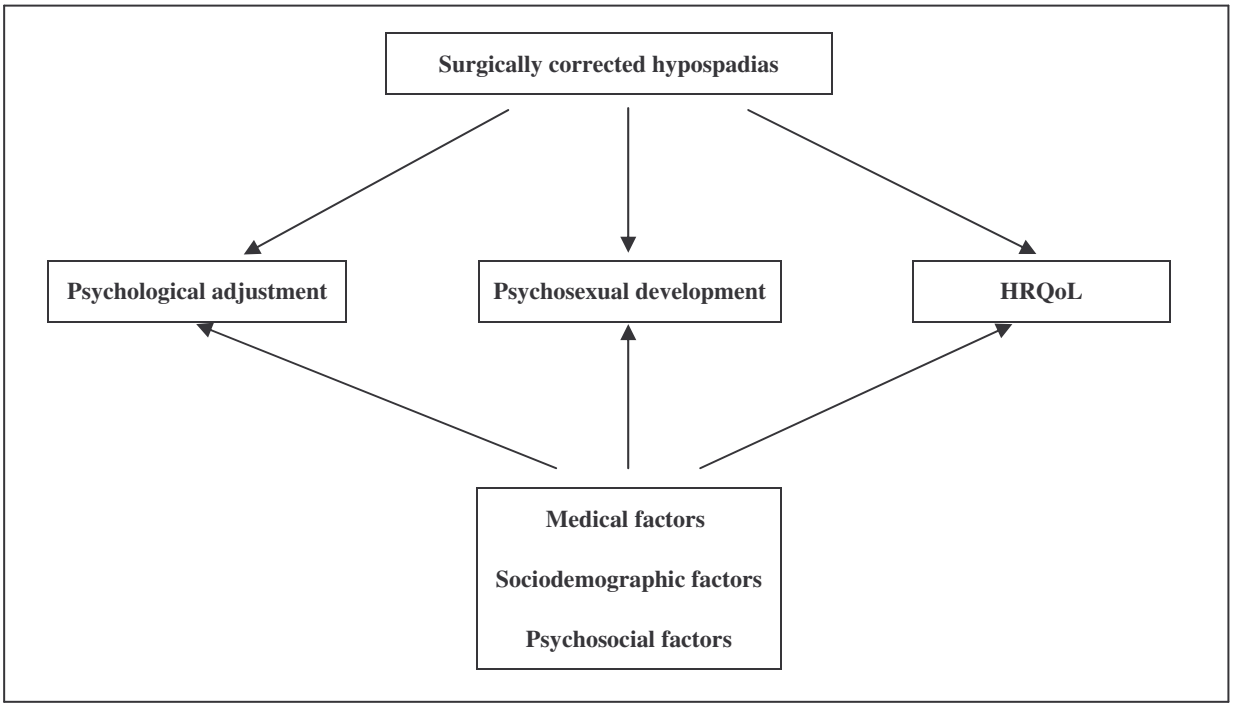

Figure 6: Schematic diagramm of the most pivotal research questions and hypotheses

1.6.2 Methods

Study design:

A cross-sectional investigation of psychological adjustment, HRQoL and psychosexual development in a large sample of boys with corrected hypospadias was conducted. Results were compared to a healthy control group.

Subjects:

The selection of patients and control subjects is described in detail in figure 7.

Patients: Children and adolescents surgically treated for hypospadias at the University Children's Hospital Zurich (1991-2005) were eligible for this study (n=147). Inclusion criteria for participation in the study were (a) age between 6 and 17 years, (b) command of the German language, (c) follow-up since last surgery ≥1 year, (d) no current or planned postoperative treatment of hypospadias, and (e) residence in Switzerland. Children with chronic diseases, disabilities or hypospadias as a concomitant phenomena in a defined disorder of sex development were excluded.

102 children fulfilled the recruitment criteria. 77 families (75.5%) agreed to participate. In 8 cases, only the child participated, whereas in 9 cases, only the parents took part. The main reason for non-participation was a lack of willingness to deal with hypospadias, stated by nine families. Further nine families confirmed their participation, but neither kept the interview appointment nor returned the questionnaire.

For demographic and medical characteristics of the sample, the reader is referred to table 3, chapter 3. No significant differences between participants and non-participants with regard to the child's age (p=.76), socioeconomic status (p=.41), nationality (p=.52) and medical characteristics such as severity of hypospadias (p=.35), age at first operation (p=.42), number of operations (p=.90), days in hospital (p=.67), and follow-up since last surgery (p=.43) were observed.

Control subjects: Each participant of the hypospadias group was matched with regard to both current age and age at first operation with a boy who was treated for an inguinal hernia. In order to have an equally large control group, 131 families had to be contacted of which 54 (41.2%) refused to participate. The main reasons for non-participation were lack of interest in the study and time consumption. Control subjects did not significantly differ from the hypospadias group with regard to nationality, socioeconomic status, age and age at first operation (see table 3, chapter 3). However, the boys with hypospadias had more operations, were hospitalized for a longer period and had their final surgery a shorter time ago. There were no differences between participating and non-participating control subjects with regard to the child's age (p=.71), socioeconomic status (p=.55), nationality (p=.95), age at first operation (p=.54), number of operations (p=.32), and follow-up since last surgery (p=.94). However, participating control subjects were hospitalized for a longer period than non-participants (p=.03).

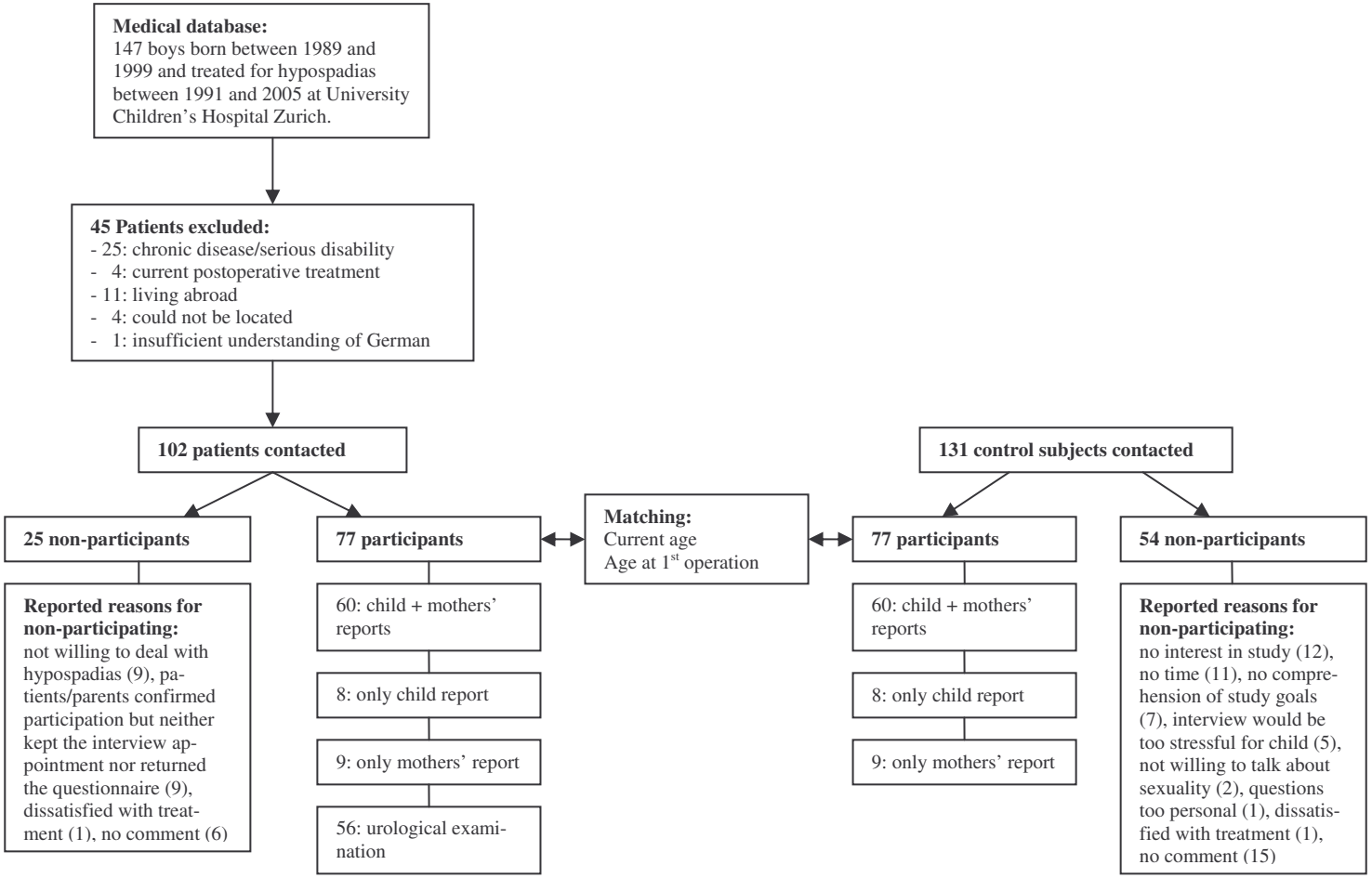

Figure 7: Flowchart of sample selection

Procedure:

The present study was approved by the local research ethics comitee. Children and parents were informed about the study by letter and were requested to return written informed consent. In case of participation, they were called in order to have a study appointment arranged. If the recipients did not respond, they were reminded of the study participation by phone. If the subjects refused to participate, they were asked for reasons and for some demographic characteristics.

The participating children and adolescents were assessed by means of a standardized face-to-face interview. Most interviews were conducted at the Children's University Hospital and lasted between 30 and 60 minutes. Participants who did not want to be subjected to the hospital's environment were visited at home. In addition, hypospadias patients were offered a physical examination by a pediatric urologist, during which standardized photographs of the penis were taken. Children's mothers were asked to answer a questionnaire at home. In one case, the father was a single parent and thus served as informant. Medical data was retrieved from the patients' records.

Children and parents who came to the hospital were refunded the travel expenses. No other reimbursements were made.

Measures:

Both the children's interview and the parents' questionnaire comprised standardized measures as well as self-developed questions and scales. The children's interview consisted of seven parts: 1) friendships, 2) HRQoL, 3) subjective concept of hypospadias, 4) experience of surgical treatment, 5) dealing with hypospadias in social situations, penile self-perception, body image, gender identity, 6) sexual experiences and sexual attitude (≥12 years), and 7) gender-role behavior (≤12 years). The mothers' questionnaire comprised eight parts: 1) sociodemographic characteristics, 2) diagnosis and attitude towards hypospadias, 3) children's HRQoL, 4) children's psychological adjustment, 5) parental attitude towards sexual development of children, 6) quality of family relationships, 7) critical life events, and 8) parental psychological adjustment.

Within the framework of the present doctoral thesis, data assessed by the following standardized measures has been published:

HRQoL:
TNO-AZL Child Quality of Life Questionnaire (TACQOL) child (CF) and parent form (PF) (Vogels et al., 2000)

Psychological adjustment:
Child Behavior Checklist (CBCL) (Achenbach, 1991)

Gender-role behavior:
Gender-role Behavior Questionnaire by Ijntema and Cohen-Kettenis (Cohen-Kettenis & Pfäfflin, 2003)

Socioeconomic status (SES):
Socioeconomic scale by Largo (developed at the University Children's Hospital Zurich)

In addition, self-developed questions and scales were applied to assess the following variables:

Penile self-perception:
Pediatric Penile Self-Perception Scale (PPPS)

Sexual attitude:
Sexual Attitude Scale

Sexual experiences:
Single questions to different sexual milestones (e. g. first french kiss) with standardized answers

Knowledge of hypospadias:
Patients were asked if they could explain why they were surgically treated on their penis (open answer).

Sense of shame due to hypospadias:
One single question with standardized answers

Experience of being teased about penile appearance by peers:
One single question with standardized answers

Mothers' concerns about sexual consequences of hypospadias:
Sum-score of three single questions with standardized answers

Mothers' general psychological stress about hypospadias:
One single question with standardized answers

The standardized measures and self-developed scales and questions are described in detail in chapters 3 and 4. Likewise, the statistical procedures applied.

The present doctoral thesis comprises the following three papers, which were all submitted to peer-reviewed journals for publication:

1) Schönbucher, V. B., Weber, D. M., & Landolt, M. A. (in press). Psychosocial adjustment, health-related quality of life and psychosexual development of boys with hypospadias: A systematic review. *Journal of Pediatric Psychology*

2) Schönbucher, V. B., Landolt, M. A., Gobet, R., & Weber, D. M. (in press). Health-related quality of life and psychological adjustment of children and adolescents with hypospadias. *Journal of Pediatrics*

3) Schönbucher, V. B., Landolt, M. A., Gobet, R., & Weber, D. M. (in press). Psychosexual development of children and adolescents with hypospadias. *Journal of Sexual Medicine*

The papers are reproduced in chapters 2, 3 and 4. Chapter 5 comprises an overall summary and discussion of the results.

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2 Psychosocial Adjustment, Health-related Quality of Life and Psychosexual Development of Boys with Hypospadias: a Systematic review.

2.1 Abstract

Objective: A systematic review of studies on psychosocial adjustment, HRQoL and psychosexual development of boys with hypospadias.

Methods: Research was conducted on several online bibliographic databases. Articles were selected on the basis of predefined criteria. Methodological quality was assessed by two independent reviewers who applied a standardized checklist. When possible, data analyses were performed by calculating effect sizes.

Results: Thirteen studies met the criteria for inclusion, whose methodological standard ranged from low to high quality. None of them has focused on HRQoL. Findings with regard to psychosocial and psychosexual adjustment were inconsistent, though they clearly showed that boys with hypospadias suffer from negative genital appraisal and sexual inhibitions. Overall, medical factors exerted a rather small influence. Psychosocial risk factors have hardly been examined so far.

Conclusions: The identification of psychosocial risk factors in methodologically sound studies is necessary to guarantee a comprehensive treatment for boys with hypospadias.

Hypospadias is the most common malformation of the penis. It is manifested by an abnormal position of the urethral opening that may lie anywhere between the glans and the scrotum. Severe forms are associated with a curvature of the penis (chordee) and a urethral opening that lies closer to the perineum. In these severe forms, surgical repair in two separate operations must be considered and complication rates tend to be higher. Explanations for the etiology of hypospadias have commonly considered genetic, endocrinological and environmental factors (Duckett, 1998). Reports on the incidence of hypospadias are inconsistent, but there is evidence that it has increased over the last decades (Dolk, 2004). Dolk estimates an incidence of approximately 3 per 1000 male births in western countries.

The surgical goal in patients with hypospadias is to achieve a typical-looking penis, to allow urination in the standing position and, particularly in severe forms of hypospadias, to enable normal sexual function. Progress in surgical techniques and pediatric anaesthesiology over the last decades now allow successful surgical repair within the first year of life (Duckett, 1998; Manzoni, Bracka, Palminteri, & Marrocco, 2004). The section on urology of the American Academy of Pediatrics has been updating its recommendations on timing for the surgery of hypospadias. In 1975 it was assumed that the period after the third year of age would be the optimal time for surgical intervention (AAP, 1975). However, in 1996 the best time for surgery to treat hypospadias was believed to be between 6 and 12 months of age. Technical considerations were no longer considered the major limiting factors in determining the timing for the surgery of hypospadias. Considerable importance was instead given to psychological factors such as emotional, cognitive and body image development that may be affected by the genital deformity and the reconstructive surgery. Superimposed on these factors, the developing sexual identity of the individual was believed to be influenced by hypospadias. It was speculated that early genital surgery could minimize disturbances in the patient's psychological development (AAP, 1996).

It is important to note that these recommendations on the timing of surgical correction are not evidence-based. Moreover, self-help groups have asked to reduce the number of surgical procedures for hypospadias because of the potential of surgical complications to cause emotional trauma (ISNA, 1994, 2006; M. Walker, personal communication, December 13, 2006). From a psychological point of view, it is indeed presumable that children and adolescents with hypospadias experience specific psychological stress which might contribute to an increased risk

of maladjustment and to an impaired quality of life. Apart from the stressors of hospitalization and genital surgery, the boys may grow up with a cosmetically and functionally-impaired penis. In addition, parental anxieties about the boys' future might result in an overprotection which may limit the patients' development of autonomy and self-confidence (APA, 1996; Berg, Berg, & Svensson, 1982; Del Priore, McHugh, Picton, & Haldane; Purschke & Standke, 1993; Sandberg et al., 2001; Walker, 1998; Zavitsanakis & Gougoudi, 2004). However, according to Bracka (1999), modern management of hypospadias is much less stressful than it used to be: "There is less physical trauma, with fewer complications, and better postoperative function and cosmesis. Hospitalizations are fewer and shorter, …." (p. 30). Therefore, it is also to be assumed that nowadays hypospadias patients grow up with fewer psychological problems than their predecessors (Bracka, 1999).

With respect to the specific development of children with hypospadias, it is important to be not only aware of the potential negative effects of the treatment and complications but also of the potential biological factors that may influence the children's behavior. There is increasing evidence that prenatal exposure to androgens does not only induce the sexual differentiation of the human genitalia but also influences postnatal sex-typed behavior patterns by engendering sex-specific brain structures. Most of the evidence comes from studies of people with disorders of sex development (DSD), which are exposed to an atypical hormone synthesis (Cohen-Bendahan, van de Beek, & Berenbaum, 2005). Jürgensen, Hiort, Holterhus, and Thyen (2007) for instance have recently found a connection between prenatal hypoandrogenization and gender-related behavior in 33 children with XY Karotypes and DSD. Although the etiology of hypospadias is still hardly intelligible, it is clearly evident that the faulty embryogenesis of the penile urethra is related to abnormalities in prenatal androgen synthesis and/or androgen receptor defects (Baskin, 2004). Therefore, Sandberg et al. (1995) hypothesized that children born with hypospadias might also tend to atypical gender-role behavior. However, this hypothesis has not been confirmed yet.

Due to the ongoing controversy about the consequences of hypospadias and its treatment, we considered it important to review systematically the research on psychosocial adjustment, HRQoL and psychosexual development of children and adolescents with hypospadias. While psychosocial adjustment incorporates a range of outcome measures including behavioral and psychosocial factors (Achenbach, 1991), HRQoL is defined as the children's health status,

weighted by the emotional response of the children themselves to their health status problems (Vogels et al., 2000). Psychosexual development, however, includes components such as the development of gender identity, gender-role behavior, body image, body inquiry, the development of sexual feelings, first sexual experiences and sexual behavior habits (Schuhrke, 1997; von Sydow, 1993). Our aim was to answer the following questions:

1. Do boys with hypospadias have an impaired psychosocial adjustment, HRQoL or psychosexual development?
2. Are there specific medical and psychosocial risk factors of the outcome?
3. How important is corrective surgery and its cosmetic outcome for the boys' adjustment?

2.2 Methods

2.2.1 Data Sources and Search Strategy

At the beginning of October 2006, a literature search was conducted for papers that focused on psychosocial adjustment, HRQoL and the psychosexual development of children and adolescents with hypospadias. The search was set up for the period between 1966 and October 2006. The following electronic bibliographic databases were searched: *Embase*, *PubMed*, *Medline*, *Cinahl*, *PsyINFO*, *Psyndex* and *Cochrane Database of Systematic Reviews*. Additionally, the databases *NDLTD*, *ProQuest Digital Dissertation* and *Dissonline.de* were searched for relevant dissertations in the field. The searches were carried out using the keywords *hypospadias*, *psychosocial*, *adjustment*, *social*, *psychological*, *adaption*, *development*, *mental health*, *behavior disorder*, *emotional stress*, *anxiety*, *personality*, *quality of life*, *psychosexual development*, *sexual satisfaction*, *sexual behavior*, *sexual dysfunction*, *sexuality, sex role, gender identity* and *follow up*. The Boolean operator *and* was used to link the search term *hypospadias* with each of the residual keywords for the psychosocial and -sexual outcome, whereas the Boolean operator *or* was employed to combine the latter with each other. This initial literature search yielded 364 articles and 83 dissertations. The *Cochrane Database of Systematic Reviews* provided 5 systematic reviews, but none of them was on the topic concerned. To augment this search result, we examined the reference lists of relevant studies and reviews. This resulted in 7 additional articles. Moreover, investigators in the field were contacted for unpublished data to minimize the problem of publication bias. This contact revealed no additional eligible data.

2.2.2 Study Selection

Firstly, titles and abstracts of articles were reviewed to screen for eligibility. Eligible publications were limited to trials that investigated psychosocial / -sexual adjustment or HRQoL of hypospadias patients in childhood and/or adolescence. Papers dealing only with the psychological consequences of hypospadias in adulthood or with medical conditions and surgical treatment were ruled out. Reviews, commentaries and studies related to other urological anomalies were also eliminated. Furthermore, 2 articles and 1 dissertation were not included because the same data were published in more than one research report.

After this initial literature review, 61 articles and 2 dissertations remained. Two authors (V.B.S., M.A.L.) read the selected articles independently from another in order to determine inclusion. When necessary, corresponding authors were contacted for additional information or clarifications of inconsistencies in the texts. Papers were included if they met the following criteria: (a) >90 % of study sample comprised individuals with hypospadias as solitary diagnosis (hypospadias as a concomitant phenomena in a defined DSD as for example Partial Androgen Insensitivity Syndrome, was not taken into account), (b) sex of participants: male, (c) mean age of participants: ≤18 years, (d) outcome: standardized self, proxy or examiner's report of psychosocial, -sexual adjustment and/or HRQoL, (e) design: case control, cross-sectional, prospective, retrospective, (f) language of publication: English, German, French. In addition, studies were excluded if they assessed sexual function or surgical outcome only, used no basic descriptive statistics (e.g. M) or lacked satisfactory quality of reporting (e.g. age at follow up missing). However, discussions were ultimately required in order to conciliate existing disagreements between the two authors. Finally, the systematic study selection led to 12 articles and 1 dissertation (figure 8).

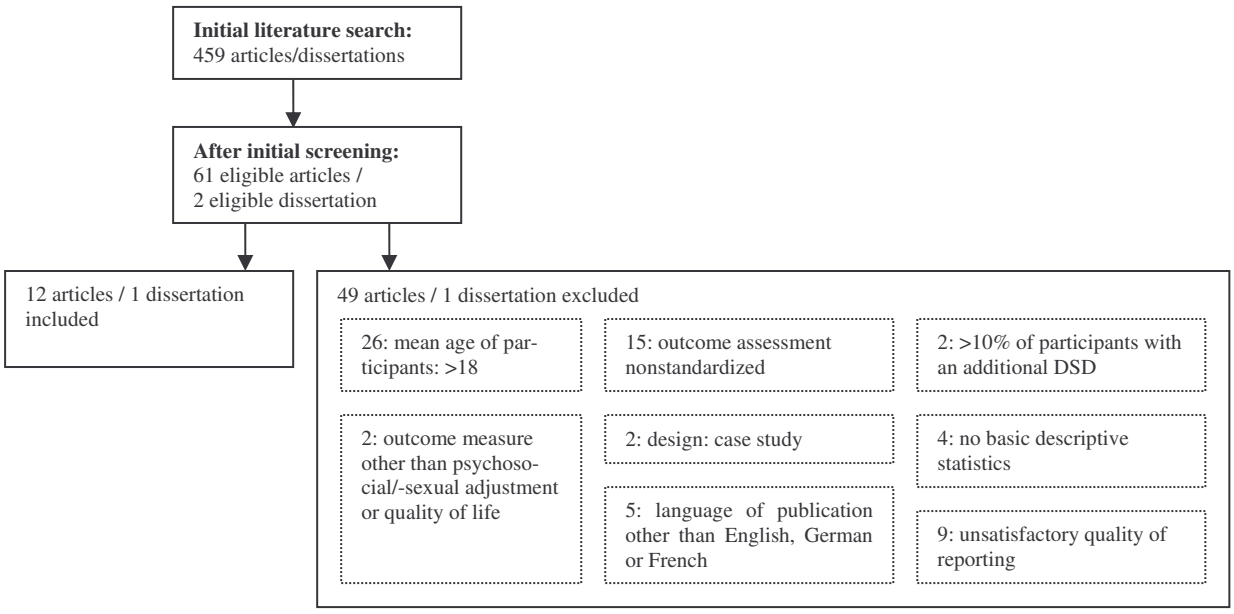

Figure 8: Flowchart of study selection. 14 articles have been excluded for more than one reason, e.g. due to nonstandardized outcome measure and mean age of participants >18. The 50 excluded studies are listed in the references and marked with two asterisks.
Note. DSD=Disorder of Sex Development

2.2.3 Assessment of Methodological Quality

Two independent reviewers (V. B. S., M. A. L.) rated the methodological quality of each included study by using a self-developed standardized checklist consisting of standards related to sample size, data selection, response rate, design, outcome assessment, statistical procedures, reporting of medical characteristics and age range of sample. Each criterion was assessed on a two-point scale (0-1) with 1 indicating a better quality. Overall scores ranged from 0 (poor) to 10 (excellent). Interrater reliability was satisfactory (Cohen's Kappa=.92; range: .71-1.00). In case of disagreement, consensus was achieved by discussion.

2.2.4 Data Analyses

In order to be able to compare the results across the different studies, we calculated effect sizes (ES) for group differences. For continuous outcomes, we calculated standardized mean differences according to Cohen (1988), using pooled standard deviations of the two groups. For dichotomous outcomes, ES were estimated based on Hasselblad und Hedges (1995). Both dichotomous and continuous outcomes were presented with 95% confidence intervals. An ES of 0.2 is considered to be small, an ES of 0.5 to be moderate, and an ES of 0.8 to be large (Cohen, 1988). A positive ES reflects a higher score in the hypospadias group, whereas a negative score reflects a higher score in the control group. Sandberg, Meyer-Bahlburg, Aran-

off, Sconzo, and Hensle (1989) presented group differences in gender-role behavior separately for five age groups. In order to summarize the data and simultaneously maintain the presentation of age effects, we combined the data into two age groups (6-8y., 9-10y.) by calculating weighted mean differences (WMD) (table 2).

However, several studies either lacked comparison data or did not report the necessary details for the calculation of ES (table 2). Furthermore, the sample of studies included in the present review was small and the outcome measures as well as the assessed constructs differed greatly across the studies (see also section "study description"). Therefore, the performance of systematic meta-analyses was considered to be inadequate. Instead, we had to narratively summarize the results but gave specific emphasize on the calculated ES. In addition, we explicitly mentioned the specific constructs or measures used in the respective study in order to mirror the heterogeneity of the assessed constructs, which are not always directly comparable (e. g. personality profile and behavior problems).

2.3 Results

2.3.1 Study Description

The main characteristics of the included studies are summarized in table 1. Four of the 13 publications focus on psychosocial adjustment, 6 on psychosexual development while 3 focus on both outcome variables. However, none of the involved studies has investigated HRQoL. The articles were published between 1989 and 2005. Four come from the United States, eight from Western Europe and one from Turkey. They were all published in English with exception of the studies by Purschke and Standke (1993) and Glaser (2004), which were both written in German. The sample sizes varied from 20 to 175 patients. Their age at follow-up ranged from 1 to 41. All of the studies but two (Mondaini et al., 2002; Purschke & Standke, 1993) incorporated only patients after corrective surgery. They were operated on at the ages of 1 month to 12 years between 1975 and 2003 (Mureau, Slijper, Slob, & Verhulst, 1997; Nelson, Bloom, Kinast, John, & Park, 2005). Except for Nelson et al., all studies included patients with both mild and severe forms of hypospadias.

Table 1: Summary of studies included in the review

Study reference/ origin	Study design/n	Age of patients (range)	Comparison data	Severity	Age at 1st op./ No. of op. (range)	Outcome measures	Method. quality (0-10)
Psychosocial Adjustment							
Eray et al., 2005 Turkey	cs 40	7-17	None	Distal to proximal	<1-14.0 1-4	CDI[a] STAIC[b]	5
Glaser, 2004 Germany	cs 208	1-41	None	Glanular to perineal	Not reported 1-2	Self-developed questions[c]	3
Mondaini et al., 2002 Italy	cs 42	18-18	500 healthy men	Glanular to perineal	Not reported 0->2	MMPI[d]	6
Mureau et al., 1997 Netherlands	cs 116	9-18	88 boys after hernia repair	Distal to proximal	<1-12 1-8	DPQ-J[e] SAS-C[f] YSR[g] CBCL[h]	9
Purschke & Standke, 1993 Germany	cs 47	5-13	23 boys after various operations norms	Glanular to penoscrotal	2-12 0-9	PFK 9-14[i] EF[j] self-developed scales[c]	4
Sandberg et al., 1989 USA	cs 69	6-10	Norms	Glanular to perineal	Not reported 1-6	CBCL[h] CBAQ[k]	7
Sandberg et al., 2001 USA	cs 175	6-10	333 healthy boys	Glanular to perineal	Not reported 1-13	CBCL[h]	7

Table 1 continued

Study reference/ origin	Study design/n	Age of patients (range)	Comparison data	Severity	Age at 1st op./ No. of op. (range)	Outcome measures	Method. quality (0-10)
Psychosexual development							
Glaser, 2004 Germany	cs 20	12-41	None	Glanular to perineal	Not reported 1-2	Self-developed questions[c]	2
Kumar & Harris, 1994 Great Britain	cs 35	13-25[l]	None	Not reported	2-12 Not reported	Self-developed questions[c]	3
Mondaini et al., 2002 Italy	cs 42	18-18	500 healthy men	Supra- to subbalanic	Not reported 0->2	Self-developed scales and questions[c]	5
Mureau et al., 1995b Netherlands	cs 116	9-18	88 boys after hernia repair	Distal to proximal	<1-12 1-8	Self-developed scales and questions[c]	7
Mureau et al., 1995a Netherlands	cs 116	9-18	88 boys after hernia repair	Distal to proximal	<1-12 1-8	Self-developed scales[m]	8
Mureau et al., 1996 Netherlands	cs 35	9-18	None	Distal to proximal	Not reported 1-8	Self-developed scales[c]	5
Nelson et al., 2005 USA	cs 43	4-26	None	Proximal	Not reported 1-2	Self-developed scales[c]	2
Sandberg et al., 1989 USA	cs 69	6-10	Norms	Glanular to perineal	Not reported 1-6	CBAQ[k] CGPQ[n]	7
Sandberg et al., 1995 USA	cs 175	6-10	333 healthy boys/norms	Glanular to perineal	Not reported 1-13	CBAQ[k] CGPQ[n] (revised)	7

Note. cs=cross-sectional; op.=operation

[a]Children's Depression Inventory. [b]State-Trait Anxiety Inventory. [c]no psychometric evaluation. [d]Minnesota Multiphasic Personality Inventory. [e]Junior Dutch Personality Questionnaire. [f]Social Anxiety Scale for Children. [g]Youth Self-Report. [h]Child Behavior Checklist. [i]Persönlichkeitsfragebogen für Kinder. [j]Encephalopathie Fragebogen. [k]Child Behavior and Attitude Questionnaire. [l]This data is based on the original medical data base, not on the final sample of participants. [m]With psychometric evaluation. [n]Child Game Participation Questionnaire.

With respect to the corresponding methodological quality, the studies considerably differed from each other (M=5.44; range: 2-9): Studies on psychosocial adjustment showed a slightly higher methodological quality (M=5.86; range: 3-9) than studies on psychosexual development (M=5.11; range: 2-8). Main weaknesses of the studies included small sample sizes, lack of comparison data, the examination of only single informants, missing information about medical characteristics, and the use of non-validated measures. The latter mainly referred to the studies on psychosexual development apart from Mureau, Slijper, Slob, and Verhulst (1995a) and Sandberg et al. (1989, 1995). Moreover, all of the publications documented cross-sectional studies. Furthermore, the operationalizations of psychosocial and psychosexual outcome differed considerably across the studies as various constructs such as behavior problems, self-confidence, social competence, personality traits, genital appraisal and gender-role behavior were assessed. It is also noticeable that most studies exclusively examined medical risk factors (table 2). The influence of psychosocial factors was investigated by just two research groups (Mureau et al., 1995a; Mureau et al., 1997; Sandberg et al., 2001).

2.3.2 Psychosocial Adjustment

Self-reports:

Results of studies and effect sizes are presented in table 2. Four studies focused on the examination of self-reported psychosocial adjustment. Mureau et al. (1997) did not report any differences in self-reported psychosocial adjustment between 116 boys with hypospadias (9-18 years) and 88 age-matched healthy control subjects on several standardized measures (DPQ-J: Junior Dutch Personality Questionnaire, SAS-C: Social Anxiety Scale for Children, YSR: Youth Self-Report). However, calculation of ES showed small significant group effects that indicate hypospadias patients to be somewhat more socially competent and to have somewhat more external behavior problems compared to the controls. The study of Mondaini et al. (2002) revealed no significant differences between 42 eighteen-year old youths with hypospadias and 500 healthy youths with regard to personality profile (MMPI: Minnesota Multiphasic Personality Inventory). Purschke and Standke (1993) evaluated the self-concepts of 47 children with hypospadias (5-13 years) by a German child personality questionnaire (PFK 9-14). Latter showed feelings of inferiority considerably more frequently but showed slightly less aggression than 23 boys of an age-matched control group who had received mild operations for various other medical problems. Yet, both hypospadias patients and the respective control group showed an equal level of (social) anxiety as well as dependence on adults.

Table 2: Major findings of studies included in the review

Study reference	Outcome	Effect sizes (95%CI)	Tested associations
		Psychosocial adjustment	
Eray et al., 2005	Not examined	Incalculable (outcome not examined)	Age at final surgery: no association
Glaser, 2004	*Self*[a]: = psychosocial adjustment[b] *Self*[a]: + emotional lability	Incalculabe (no comparison data)	Not examined
Mondaini et al., 2002	*Self*: = personality profile	0.24** (-0.08-0.56)	Severity: no association No. of operations: no association Age at final surgery: no association
Mureau et al., 1997	*Self:* = social inadequacy *Self:* = self-confidence *Self:* = social anxiety *Self:* = social competence *Self:* = behavior problems external internal total *Proxy:* = behavior problems external internal total	0.15* (-0.12-0.43) 0.19* (-0.09-0.47) 0.00* (-0.28-0.28) 0.29* (0.01-0.57) 0.29* (0.01-0.57) 0.02* (-0.26-0.30) 0.18* (-0.10-0.46) -0.07* (-0.35-0.21) -0.08* (-0.35- -0.20) -0.02* (-0.29-0.26)	Severity: no association Surgical procedure: no association No. of operations: no association Age at final surgery: no association Age at follow-up: no association Genital perception: pos. association with self-confidence neg. association with behavior problems Body image: neg. association with social anxiety neg. association with behavior problems
Purschke & Standke, 1993	*Self:* = anxiety *Self:* = aggressiveness *Self*: = social anxiety *Self:* = dependence on adults *Self:* + feelings of inferiority *Proxy:* + behavior problems	-0.17** (-0.67- -0.33) -0.38** (-0.88- -0.12) 0.20** (-0.30-0.70) 0.13** (-0.37-0.63) 0.98** (0.46-1.51) Incalculable (missing information)	Severity: pos. association with behavior problems no association with self-reported psychological adjustment No. of operations: pos. association with behavior problems neg. association with self-reported psychological adjustment Age at 1st operation: ideal age 5-6 years Surgical outcome: inconsistent findings

Table 2 continued

Study reference	Outcome	Effect sizes (95%CI)	Tested associations
		Psychosocial adjustment	
Sandberg et al., 1989	*Proxy:* + behavior problems		Severity: partial positive associations with behavior problems
	external	0.21* (-.06-.47)	negative associations with academic achievement
	internal	0.29* (0.03-0.55)	no association with social competency
	total	0.33* (0.07-0.59)	No. of hospitalizations: inconsistent findings
	Proxy: – social competence	-0.29* (-0.55- -0.03)	
	Proxy: = academic achievement	0.10* (-0.16-0.36)	
Sandberg et al., 2001	*Proxy:* – behavior problems		Severity: no association
	external	-0.36* (-0.55- -0.18)	No. of operations: partial pos. associations with behavior problems
	internal	Incalculable (missing information)	No. of hospitalizations: partial pos. associations with behavior problems
	total	Incalculable (missing information)	Parents genital appraisal: pos. association with academic achievement
	Proxy: – social competence	0.00* (-0.18- -0.18)	
	Proxy: = academic achievement	Incalculable (missing information)	
		Psychosexual development	
Glaser, 2004	*Self:* = masturbation frequency	Incalculable (no comparison data)	Not examined
	Self: = intercourse-frequency		
Kumar & Harris, 1994	*Self:* – genital perception	Incalculable (no comparison data)	Not examined
	Self: – experience of sex. intercourse		
Mondaini et al., 2002	*Self:* + sex. inhibitions	0.60** (0.28-0.92)	Severity: neg. association with genital appraisal
	Self: – experience of sex. intercourse	-0.71** (-1.03- -0.39)	No. of operations: most sexual inhibitions if not operated on
	Self: – genital perception	1.57** (1.24-1.90)	Age at final surgery: no association
Mureau et al., 1995b	*Self:* = age at 1st sex. experiences[c]	-0.12* (-0.40-0.16) - 0.36* (0.09-0.64)	Severity: no association with sexual inhibitions
	Self: = sex. behavior[d]	-0.09* (-0.36-0.19) - 0.22* (-0.06-0.49)	Surgical procedure: no association
	Self: + sex. inhibitions	1.53** (1.17-1.89)	No. of operations: no association
	Self: – genital perception	Incalculable (missing information)	Age at final surgery: no association
			Age at follow-up: neg. association with genital perception
			neg. association with sexual inhibitions

Table 2 continued

Study reference	Outcome	Effect sizes (95%CI)	Tested associations
		Psychosocial adjustment	
Mureau et al., 1995a	*Self:* – genital perception *Self:* = body image	-0.52* (-0.80- -0.24) 0.19* (-0.09-0.47)	Severity: no association No. of operations: no association Surgical procedure: no association Age at final surgery: no association Age at follow-up: neg. association with body image Response from others: no association
Mureau et al., 1996	*Self:* – genital perception[e]	-0.86* (-1.35- -0.37)	Penile length: no association
Nelson et al., 2005	*Self:* – genital perception	Incalculable (no comparison data)	Not examined
Sandberg et al., 1989	*Proxy:* + feminine behavior *Proxy:* = masculine behavior	6-8y.: 0.18* (0.10-0.26) - 0.58* (0.50-0.66)[f] 9-10y: -0.24* (-0.34- -0.14) - 0.69* (0.59-0.79)[f] 6-8y: 0.21* (0.13-0.29) - 0.37* (0.29-0.45)[f] 9-10y: -0.44* (-0.54- -0.34) - 0.58* (0.48-0.68)[f]	Severity: no association No. of hospitalizations: no association
Sandberg et al., 1995	*Proxy:* = feminine behavior *Proxy:* + masculine behavior	-0.26* (-0.45-0.08) - 0.26* (0.08-0.45)[f] 0.17* (-0.02-0.35)	Severity: no association No. of hospitalizations: pos. association with feminine behavior

Note. – indicates lower scores compared to controls/norms or indicating low scores if controls/norms are missing; + indicates higher scores compared to controls/norms or indicating high scores if controls/norms are missing; = indicates missing difference between cases and controls/norms or indicating normal scores if controls/norms are missing; * ES was calculated according to Cohen (1988) due to continous outcome; ** ES was calculated according to Hasselblad & Hedges (1995) due to dichotomous outcome; self=self-report measures; proxy=proxy-report measures; sex=sexual

[a]If patients were too young for the children's interview, Glaser assessed psychosocial adjustment by means of proxy-report; [b]e. g. sleeping problems, anxiety, separation anxiety; [c]e. g. age at first being in love, at first kiss, at first sexual intercourse; [d]e. g. sexual desire, sexual activity, sexual satisfaction; [e]compared to surgeon's perception; [f]dependent on the specific subscale

Likewise, Glaser (2004)[1] presented inconsistent findings: Whereas the twenty hypospadias patients showed a high level of emotional instability (unvalidated questions; ES not calculable), Glaser did not notice any further impairments of psychosocial adjustment (e. g. an increased level of separation anxiety).

The inconsistent results on self-reported psychosocial adjustment (range ES's:-.038-0.98) might be a result of their different methodological standard. Studies with a higher standard (Mondaini et al., 2002; Mureau et al., 1997) suggested a better psychosocial adjustment. However, other differences between the studies that were related not only to their origins and the subjects' ages but also to the diversity of applied measures could also have caused the contradictory results.

Proxy-reports:

Mostly, proxy-reported psychosocial adjustment was measured with the Child Behavior Checklist (CBCL) (Mureau et al., 1997; Sandberg et al., 1989, 2001). However, Sandberg et al. (1989) additionally used the Behavior Disturbance Scale of the Child Behavior and Attitude Questionnaire (CBAQ), while Purschke and Standke (1993) used the German "Encephalopathie-Fragebogen (EF)" as well as unvalidated questions (table 1). The results of proxy-reports are also inconsistent (table 2). The study of Mureau et al. (1997) showed that the parents of 116 boys with hypospadias did not report their children's behavior to be more problematic compared to the parents of 88 age-matched controls, treated for an inguinal hernia. Conversely, the findings of Purschke and Standke (1993) suggest that the 47 children with hypospadias (5-13 years) had more behavior problems than the 23 children of the same age, who had been subjected to a variety of mild operations for other medical problems (ES not calculable). In 1989, an American research group (Sandberg et al.) found more behavior problems and lower social competence in 69 children with hypospadias (6-10 years) compared to the corresponding norms of 300 healthy children between the ages of 4 to 16 years. Calculated ES were significant but small. Twelve years later, the same researchers (Sandberg et al., 2001) published a study with a slightly better methodological standard that involved a larger sample of hypospadias patients (n=175) as well as a control group of healthy school boys (n=333). Although the calculated ES of mean difference suggests no group effect, multiple regression[2], controlling for sociodemographic characteristics, showed that the parents of hy-

[1] If patients were too young to attend the children's interview, Glaser assessed psychosocial adjustment by means of proxy-report

[2] Effect sizes for multivariate analyses could not be calculated due to missing information

pospadias patients (6-10 years) considered their children to be less socially competent than the parents of the comparison subjects (6-10 years). Yet, in contrast to their earlier results, boys with hypospadias were judged by their parents to have fewer externalizing behavior problems compared to the parents of the controls (ES small to moderate).

Regarding the inconsistencies of proxy-reports on psychosocial adjustment, it is important to take the studies' methodological quality into account. Comparable with the findings of self-reports, studies with a methodologically higher qualified standard (Mureau et al., 1997; Sandberg et al., 2001[3]) suggested a somewhat better psychosocial adjustment. Calculated ES', however, tended to be small and varied less (range: -0.29-0.33) than ES's for self-reports. Furthermore, there are other differences between the studies (e. g. age range of subjects) that might be responsible for the contradictory findings.

Risk factors for psychosocial maladjustment:

Medical factors: Except for Glaser (2004), all studies on psychosocial adjustment have focused on the association with medical risk factors (table 2). The results indicate that the association between psychosocial adjustment and medical factors is weak. According to Mureau et al. (1997) and Mondaini et al. (2002), psychosocial adjustment (measured with several standardized outcome measures) and personality profile (MMPI) were not related to the severity of hypospadias, the number of operations and the age at final surgery. Neither did the type of surgical procedure correlate with psychosocial adjustment (Mureau et al., 1997). Moreover, Mondaini et al. (2002) did not detect any difference in personality profile between treated and untreated patients. Additionally, a recent study by Eray et al. (2005) showed that patients whose corrective surgery was completed before the age of eighteen months did not differ in their level of depression and anxiety from patients who had their final operation at a later age. However, the findings of Purschke and Standke (1993) and Sandberg et al. (1989, 2001) did not coincide with those of the above mentioned authors. Namely, Purschke and Standke reported that patients with a more severe form of hypospadias and who were surgically treated more often showed a higher incidence of behavior problems. In addition, the number of operations was negatively associated with the patients' self-concept. However, the eight untreated patients displayed the lowest psychosocial adjustment. The quality of surgical

[3] Sandberg et al. (2001) did not show a higher score in our methodological assessment than Sandberg et al. (1989), but the former involved a larger sample as well as a healthy control group and used revised measures for the assessment of gender-role behavior, which showed more favorable psychometric criteria. However, we did not consider this fact in our methodological rating due to practical reasons.

outcome correlated negatively with behavior problems but positively with the patient's psychological strain. Furthermore, children operated on between the ages of five and six showed fewer behavior problems than children operated on either between the ages of two and four or between six and twelve. The results of the two studies of Sandberg et al. (1989, 2001) are, however, inconsistent. While the first study suggested that children with a more severe form of hypospadias showed more behavior problems and better school achievement (CBCL), the second investigation did not confirm this assumption. However, in 2001 the researchers detected a positive association between the patients' behavior problems and the number of operations as well as the number of hospitalizations the children had been subjected to.

Psychosocial factors: According to Mureau et al. (1997), boys who were more satisfied with the appearance of their genitals and their overall body image had fewer behavior problems, were more self-confident and showed less social anxiety. In addition, the subjects' age was not found to be associated with psychosocial adjustment. Sandberg et al. (2001) reported that the parents' satisfaction with the appearance of the patients' genitals were positively correlated with their academic achievement but not so with their behavior problems.

2.3.3 Psychosexual Development

Self-reports:

Seven articles focus on at least one aspect of self-reported psychosexual development (Glaser, 2004; Kumar & Harris, 1994; Mondaini et al., 2002; Mureau et al., 1995a, 1995b; Mureau, Slijper, Slob, Verhulst, & Nijman, 1996; Nelson et al., 2005), three of which are based on the same investigation (Mureau et al.) (table 2). The most frequently examined construct is genital perception that has been assessed either as the satisfaction with genital appearance or as the appraisal of one's genitals to look normal. Apart from Mureau et al. (1995a), all authors used non-validated self-constructed measures. Authors reported between 20 and 80 % of the boys with hypospadias suffered from negative genital perception, while healthy age-matched controls were significantly more satisfied with penile appearance (Kumar & Harris, 1994; Mondaini et al., 2002; Mureau et al., 1995a, 1995b). ES's were moderate (Mondaini et al., 2002) to large (Mureau et al., 1995a (table 2). Interestingly, Mureau et al. (1996) did not detect any relation between the patients' and the surgeon's perception of genital appearance. Overall, the latter was more satisfied, indicated by a large ES.

With regard to sexual inhibitions, both Mondaini et al. (2002) and Mureau et al. (1995b) found that children and adolescents with hypospadias reported more sexual inhibitions than the healthy age-matched controls (moderate to large ES's). They were not only more afraid of being rejected by a potential partner due to genital appearance but also prone to hide genitals in public lavatories (Mondaini et al., 2002; Mureau, 1995b). However, the available studies do not clarify whether youths with hypospadias also show a delay of first sexual experiences. In the methodologically most sound study (Mureau et al., 1995b), boys with hypospadias (9-18 years) had their first sexual experiences (e. g. first kiss) at slightly higher ages than the age-matched controls treated for an inguinal hernia, except for masturbating. Yet, ES's were only small (table 2). In the study of Mondaini et al. (2002), however, the calculated ES indicates that adolescents with hypospadias have their first sexual intercourse at a considerably higher age than healthy boys. Kumar and Harris (1994) reported the same, although they did not include a control group. However, results indicate that adolescents with hypospadias do not differ from healthy youths neither with regard to sexual activity, number of coitus partners and sexual desire, nor with regard to body image (Glaser, 2004; Mureau et al., 1995a, 1995b).

Proxy-reports:

Sandberg et al (1989, 1995) examined proxy-reported gender-role behavior measured by the gender-role behavior scales of the Child Behavior and Attitude Questionnaire (CBAQ) and by the Child Game Participation Questionnaire (CGPQ) (table 1). Their findings were, however, contradictory (table 2). In 1989, consistent with a hormonal hypothesis of gender-role development, they observed more feminine behavior in 6-8 year old boys with hypospadias (6-10 years) than in a reference group of healthy male children. Calculated ES are small to moderate, depending on the specific subscale. Yet, 6-8 year old children with hypospadias showed also slightly higher scores with regard to masculine behavior patterns, indicated by the small significant ES. Even more inconsistent were the findings with regard to the 9-10 year old patients, with ES ranging from negative in the moderate realm to positive in the large realm. Nevertheless, the authors reported that half of the boys even showed similar levels of feminine behavior to a reference group of 23 gender-dysphoric children (ES not calculable due to missing information). Six years later, they published a second study that involved revised versions of the CBAQ and CGPQ. Although calculated ES indicate somewhat inconsistent findings, they suggest that the behavior patterns of 175 children with hypospadias did not considerably differ from that of the 333 boys of a healthy control group. Moreover, multivari-

ate analyses[2] showed that sociodemographic control variables were stronger predictors of gender-role behavior than genital status.

Risk factors for psychosexual development:

Medical factors: The relationship between medical characteristics and the psychosexual development of boys with hypospadias was examined by three research groups (Mondaini et al., 2002; Mureau et al., 1995a, 1995b; Sandberg et al., 1989, 1995). Overall, the findings imply that the association between medical characteristics and psychosexual development is rather weak (table 2). However, Mondaini et al. (2002) reported a negative effect of severity of hypospadias on genital appraisal (non-validated), Mureau et al. (1995b) found a positive effect of severity on sexual inhibitions (non-validated) and Sandberg et al. (2001) detected a positive effect of number of hospitalizations on feminine behavior patterns (CBAQ, CGPQ). Furthermore, Mondaini et al. (2002) reported that untreated adolescents with hypospadias had more sexual inhibitions (non-validated) than treated boys. However, no association was found between psychosexual development and number of operations, age at final surgery as well as type of surgical procedure.

Psychosocial factors: Mureau et al. (1995b) reported that the boys' age was negatively related to genital perception and positively associated with sexual inhibitions. Conversely, another study by Mureau et al. (1995a) showed that age was negatively related to body image but not to genital perception. This inconsistency between the studies is assumed to be a result of the divergent assessments of genital perception. In addition, the experience of having been teased due to genital appearance was not linked to genital perception.

2.4 Discussion

We conducted the first systematic review of studies on psychosocial adjustment, psychosexual development and HRQoL of children and adolescents with hypospadias. Furthermore, we aimed to assess psychosocial and medical risk factors of the outcome as well as the psychological significance of corrective surgery and its cosmetic result.

Thirteen studies met the criteria for inclusion, whose methodological standard ranged from low to high quality. All studies focused on either psychosocial adjustment or psychosexual development. With regard to psychosocial adjustment, the results of the studies are inconsistent, with recent and methodologically more sound studies indicating a slightly more favorable outcome (Mondaini et al., 2002; Mureau et al., 1997; Sandberg et al., 2001). The inconsistent results may be a consequence of the variation in methodological quality. On the other

hand, surgical treatment strategies have evolved over the last decades: Patients are being operated at a younger age, surgical techniques and anesthesia have been improved and nursing practice has adapted to children's needs as for example by the rooming-in of parents (Bracka, 1999). These varying standards of treatment make direct comparisons of results difficult and may be responsible for a better psychosocial adjustment in recent studies. Yet, it is likely that the differences between the studies with regard to origin, age range of subjects and operationalization of psychosocial adjustment contributed to contradictory findings.

The findings regarding psychosexual development, however, clearly demonstrate that boys with hypospadias suffer from negative genital appraisal and sexual inhibitions. Nevertheless, age at first sexual experiences and general sexual behavior appeared to be less affected by their penile malformation (Kumar & Harris, 1994; Mondaini et al., 2002; Mureau et al., 1995a, 1995b; Nelson et al., 2005). The effects of hypospadias on gender-role behavior, however, are not clarified yet (Sandberg et al., 1989, 2001). Thus, the hypothesis that prenatal hypoandrogenization associated with hypospadias interferes with the development of gender-typical masculine behavior remains speculative and thus requires further examination.

The findings on the significance of medical factors are likewise inconsistent. Overall, they suggest that medical characteristics bear a rather small influence on the psychosocial and psychosexual outcome. It is important to note that there is no empirical evidence that corrective surgery at the youngest possible age leads to a better psychological development. Thus, empirical results do not support the early surgical interventions, which pediatric urologist recommend for (APA, 1996; Zavitsanakis & Gougoudi, 2004).

Evidence with regard to psychosocial predictors of adjustment is scarce. The results of Mureau et al. (1997) suggest that positive genital perception and a healthy body image are crucial for the boys' psychosocial adjustment. However, it is also possible that psychosocially better adjusted children develop a more favorable genital and body perception. In addition, there is some evidence that genital appraisal of the patients' parents is connected to the patients' adjustment (Sandberg et al., 2001), which indicates that parental attitude towards hypospadias might be important for the patients' well-being. However, the findings with regard to the significance of the children's current age are contradictory (Mureau et al., 1995a, 1995b, 1997). Yet, it would be plausible if adolescents were less satisfied with penile appearance than younger boys due to a change in physical self-perception as well as the increasing importance of sexuality during puberty.

Finally, no conclusion can be drawn with regard to the importance of corrective surgery itself for the children's psychosocial and psychosexual development. Although Purschke and Standke (1993) and Mondaini et al. (2002) reported some differences in psychosocial and psychosexual adjustment between untreated and treated boys, the examined groups of untreated patients were much too small (n=6 and n=8) to generalize results.

2.4.1 Limitations and Directions for Future Research

There are several factors that limit the generalization of the findings. Firstly, many studies lack methodological quality due to small sample sizes, the absence of control groups or norms and the use of non-validated measures, particularly with respect to psychosexual outcome. Secondly, the number of existing studies is too small to draw general conclusions. Ultimately, it is assumed that modern management of hypospadias is much less stressful than it used to be (Bracka, 1999): Children with hypospadias are currently operated at a younger age with more sophisticated surgical techniques which are bound to generate a better surgical outcome.

Other limitations relate to research questions which have yet to be investigated. Surprisingly, there exists no study on HRQoL of children with hypospadias. HRQoL measurement has emerged as an important tool for analyzing health outcome in clinical trials (Eiser & Jenney, 2007). It is a multidimensional construct defined as the patient´s perception of the effects of illness/disability and treatment on his physical, psychological and social funcions. It is thus not solely confined to physiological outcome and/or psychopathological symptoms (Matza, Swensen, Flood, Secnik, & Leidy, 2004). Both generic and disease-specific HRQoL-measures have been developed. While the former allows comparisons across patient groups as well as with healthy children, the latter appear to be more sensitive to the implications of a single condition (Eiser & Jenney, 2007). Furthermore, it is recommended to make both self- and proxy-ratings when measuring HRQoL: both the child's and the parents's perspective should be assessed (Matza et al., 2004). One tool that provides both a patient's and a parent's form as well as the combination of a generic measure with disease-specific moduls is the Pediatric Quality of Life Questionnaire (PedsQL) (Varni, Seid, & Rode, 1999). However, the PedsQL does not comprise a hypospadias-specific modul, which would allow a more detailed assessment of HRQoL in children with hypospadias. Unfortunately, to the best of our knowledge a HRQoL measurement that is sensitive to the specific challenges of hypospadias does not exist and has yet to be developed. Ideally, it would assess the child's perception of his physical (e. g. micturition, sexual function), psychological (e. g. depressive emotions, senses of shame,

body image) and social (e. g. social withdrawal, sexual anxieties, being teased by peers, concealment of hypospadias) functions.

Likewise, there is a lack of knowledge about the influence of psychosocial factors on the patients' development. It is presumable that family background variables (e.g. the quality of family relationships), the patients' coping behavior as well as the attitude of patients' parents towards their children's penile condition may have a greater impact on the boys' psychological development than medical characteristics. Another direction for future research would be an extended examination of the psychosexual development. The majority of existing studies examined only a certain aspect of psychosexual development. However, the construct of psychosexual development includes many components such as the development of gender identity, gender-role behavior, body inquiry and the development of sexual feelings (Schuhrke, 1997; von Sydow, 1993). Thus, it is important to note that the psychosexual development is not exclusively related to first sexual experiences or to sexual behavior habits. Finally, it would be interesting to shed light on the psychological development of untreated children with hypospadias in order to be able to investigate the psychological consequences of hypospadias repair. This aspect should be raised since, on the one hand, self-help groups have recently announced the need for a reduction in the frequency of genital surgery due to the potential of emotional trauma (ISNA, 2006; M. Walker, personal communication, December 13, 2006) and, on the other hand, pediatric urologists plead for surgical correction of hypospadias in order to prevent negative repercussions on the patients' psychosexual adjustment (AAP, 1996; Zavitsanakis & Gougoudi, 2004).

A final limitation refers to the methodical procedures by which we have conducted the present systematic review. Although we have applied systematic review techniques to select relevant articles and have provided a reliable rating of methodological quality, the chosen sample of studies did not allow the conduction of systematic meta-analyses. Consequently, a sophisticated examination of trends in data, their relations to methodological quality and specific outcome measures as well as analyses of potential differences between self- and proxy-report could not be performed. Nevertheless, by calculating effect sizes we tried to reduce subjectivity in the narrative presentation of the results.

2.4.2 Clinical Implications

The inconsistent results and the lack of knowledge about the influence of psychosocial factors on the psychological development of boys with hypospadias allow only limited implications for clinical practice. Based on the findings that boys with hypospadias tend to suffer from negative genital appraisal more often than healthy boys, one may assume that they might profit from psychosocial support for the development of sufficient self-confidence which would possibly encourage them to better accept their penis. Surgeons must be aware that genital appraisal does not exclusively rely on the quality of surgical outcome (Mureau et al., 1996). Thus, optimal medical care for hypospadias patients cannot merely be achieved by optimizing surgical results. In order to guarantee a comprehensive treatment concept, the incorporation of psychological factors such as the anxiety of rejection by eligible sex partners may be equally important. Moreover, it is tenable that genital appraisal and sexual inhibitions may be affected to a higher degree during adolescence than during childhood. Hence, it is believed to be essential, as already Berg, Berg, Svensson, and Aström (1981) argued twenty-six years ago, that patients with hypospadias will need to be followed-up until young adulthood. Apart from regular urological medical examinations, the patients should be appropriately informed about their penile condition and to be offered psychological support.

In summary, the present review demonstrated that there is a lack of evidence regarding psychosocial adjustment, psychosexual development and HRQoL of children and adolescents with hypospadias. Furthermore, the review pointed out that the guidelines for surgical treatment are partly based on psychological assertions that have not been empirically confirmed. Particular attention should be paid to the assumption that a surgical correction of hypospadias at the earliest age possible leads to a minimization of adverse effects on the boy's development. From a psychosocial point of view, an optimization of the treatment of boys with hypospadias ultimately requires a clarification of the patient's psychosocial and psychosexual development in methodologically sound investigations.

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3 Health-Related Quality of Life and Psychological Adjustment of Children and Adolescents with Hypospadias

3.1 Abstract

Objectives: To assess health-related quality (HRQoL) of life and psychological adjustment of children and adolescents after hypospadias repair and to identify sociodemographic, medical and psychosocial predictors of outcome.

Study design: Seventy-seven boys (7-17 years) operated on for hypospadias were investigated in a cross-sectional study. Child- and mother-rated HRQoL was evaluated by the *TNO-AZL Child Quality of Life Questionnaire*, psychological adjustment by the Child Behavior Checklist. Scores were compared to an age-matched control group consisting of 77 boys after hernia repair.

Results: Compared to the controls, self-reported HRQoL of hypospadias patients was lower in most dimensions. In contrast, mother-reported HRQoL and psychological adjustment did not differ between the two groups. Higher age and "being less ashamed of penile appearance" predicted better self-reported HRQoL, while positive penile self-perception, the patient's experience of not having been teased about his penis and more severe hypospadias contributed to better mother-reported HRQoL. Psychological adjustment was predicted by higher age, Swiss nationality, more severe hypospadias and the patient's experience of not having been teased about his penis.

Conclusions: In contrast to psychological adjustment, self-reported HRQoL of boys with hypospadias can be diminished. Psychosocial factors are more important predictors than medical variables.

Hypospadias is the most common malformation of the penis with a reported incidence ranging up to 3 per 1000 male births (Dolk, 2004). It is manifested by an abnormal position of the urethral opening, which lies anywhere between the glans and the scrotum. In most hypospadias, the prepuce is split and severe forms are associated with a curvature of the penis (chordee).

The goal of hypospadias repair is to achieve a normal looking penis, to allow micturition in the standing position and to enable full sexual functions (Duckett, 1998). Although it is commonly assumed that both genital surgery and residual cosmetic or functional deficits may cause psychological stress to the patients (Bracka, 1999; Mureau, Slijper, Slob, & Verhulst, 1997), only few studies report on the patients' psychological adjustment during childhood and adolescence (Schönbucher, Weber, & Landolt, in press). Furthermore, findings are inconsistent: In older and methodologically less sound studies, children with hypospadias showed increased rates of psychological maladjustment (Purschke & Standke, 1993; Sandberg, Meyer-Bahlburg, Aranoff, Sconzo, & Hensle, 1989), whereas outcome is better in more recent, methodologically sound studies (Mondaini et al., 2002; Mureau et al., 1997; Sandberg et al., 2001). Previous findings on the significance of medical factors for psychological adjustment are inconsistent too. However, most authors suggest that medical characteristics such as severity of hypospadias, number of operations and age at first and final surgery bear a rather small influence on the patient's psychological adjustment (Eray et al., 2005; Mondaini et al., 2002; Mureau et al., 1997; Purschke & Standke, 1993; Sandberg et al., 2001). In spite of these weak associations between medical factors and psychological adjustment, psychosocial risk factors have hardly been examined.

Moreover, no studies on health-related quality of life (HRQoL) of boys with hypospadias have been published. This is striking because it is known from children with other deformities such as bladder extrophies or anorectal malformations that their quality of life can be impaired (Poley, Stolk, Tibboel, Molenaar, & Busschbach, 2004; Stjernqvist & Kockum, 1999). The multidimensional concept of HRQoL is defined as the patient's subjective evaluation of his/her physical, cognitive, social and emotional functioning. It has recently become established as one of the most important approaches in assessing significant consequences of the patients' state of health (Edwards, Patrick, & Topolski, 2003).

This study aimed at a comprehensive, cross-sectional investigation of both HRQoL and psychological adjustment, comparing a large sample of boys operated on for hypospadias and a healthy control group. We hypothesized that boys with hypospadias would show lower HRQOL and more psychological adjustment problems than the controls. Furthermore, we postulated that sociodemographic, medical and psychosocial factors are important predictors of the outcome.

3.2 Methods

3.2.1 Subjects

Hypospadias group:

Children and adolescents operated on for hypospadias at the University Children's Hospital Zurich (1991-2005) were eligible for this study (n=147). Inclusion criteria for participation in the study were (a) age between 6 and 17 years, (b) command of the German language, (c) follow-up since last surgery ≥1 year, (d) no current or planned postoperative treatment of hypospadias, and (e) residence in Switzerland. Children with chronic diseases, disabilities or hypospadias as a concomitant phenomena in a DSD were excluded.

102 children fulfilled the recruitment criteria. 77 families agreed to participate (75.5%). In 8 cases, only the child participated, whereas in 9 cases, only the mothers took part. Demographic and medical characteristics of the sample are listed in table 3. Most frequent complications leading to further surgery were fistulas and stenoses. No significant differences were observed between participants and non-participants with regard to the child's age (p=.76), socioeconomic status (p=.41), nationality (p=.52) and medical characteristics such as severity of hypospadias (p=.35), age at first operation (p=.42), number of surgeries (p=.90), days in hospital (p=.67), and follow-up since last surgery (p=.43).

Control group:

Each participant of the hypospadias group was matched with regard to both current age and age at first operation with a boy who was treated for an inguinal hernia. In order to have an equally large control group, 131 families had to be contacted of which 54 (41.2%) refused to participate. Control subjects did not significantly differ from the hypospadias group with regard to nationality, socioeconomic status, age and age at first operation (table 3). However, the boys with hypospadias had more surgeries, were hospitalized for a longer period and had their final surgery a shorter time ago.

Table 3: Demographic and medical characteristics of patients and controls

	Boys operated for hypospadias (n=77)	Boys operated for inguinal hernia (n=77)	p†
Age			
Mean (SD)	11.03 (3.31)	11.34 (3.32)	.52
Median (Range)	9.97 (6.66-17.34)	10.69 (6.80-18.23)	
Nationality (%)			
Swiss	60.8	71.6	.16
Foreigners*	39.2	28.4	
Socioeconomic status (%)			
Lower	23.3	15.1	.57‡
Middle	47.9	60.3	
Upper	28.8	24.7	
Age at 1st operation			
Mean (SD)	3.01 (2.37)	2.83 (2.77)	.19
Median (Range)	2.20 (0.20-11.87)	1.90 (0.16-14.18)	
No. of hypospadias/hernia surgeries			
Mean (SD)	1.86 (2.06)	1.09 (0.29)	<.0001
Median (Range)	1.00 (1.00-15.00)	1.00 (1.00-2.00)	
Days in hospital			
Mean (SD)	11.59 (12.67)	2.68 (1.16)	<.0001
Median (Range)	8.00 (1.00-77.00)	2.00 (1.00-7.00)	
Follow-up since last surgery			
Mean (SD)	6.82 (3.51)	8.40 (3.66)	.002
Median (Range)	6.58 (1.00-15.15)	7.68 (0.80-16.13)	
Severity of hypospadias (%)			
glanular	18.2	-	-
coronal	35.1	-	-
subcoronal	9.1	-	-
penil	33.8	-	-
penoscrotal	3.9	-	-

Note: †U-Tests according to Mann-Whitney for continuous variables, χ^2-Tests for categorical variables; ‡ U-Test according to Mann-Whitney based on SES scores; *mainly originating from former Yugoslavia

3.2.2 Procedure

The present study was approved by the local research ethics committee. Children and parents were informed about the study by letter and were requested to return a written informed consent form. The participating children and adolescents were assessed by means of a standardized interview, which was usually conducted at the hospital. In addition, hypospadias patients were offered a physical examination by a pediatric urologist, during which standardized photographs of the penis were taken. Patients' mothers were asked to answer a questionnaire at home.

3.2.3 Measures

Health-related quality of life:

The TNO-AZL Child Quality of Life Questionnaire (TACQOL) is a generic instrument for measuring HRQoL (Vogels et al., 2000). It consists of both a child form (CF) and a parent form (PF). The TACQOL contains 5 health scales and 2 emotional scales, each consisting of 8 items: physical functions, motor functions, autonomy, cognitive functions, social functions, global positive emotional functions and global negative emotional functions. Maximum domain scores are 32 for the health scales and 16 for the emotional scales. Higher scores correspond to better HRQoL. To obtain a reliable measure of overall quality of life, we computed a total score by summing the items across all scales (maximum score: 192 points). The TACQOL has been validated in several patient groups and a community sample of healthy Dutch children with Cronbach's α ranging from .67 to .84 (Vogels et al., 2000).

Psychological adjustment:

The Child Behavior Checklist (CBCL) is a 120-item standardized parent-report measure of behavior problems in children (Achenbach, 1991). Higher scores indicate a greater number or intensity of behavior problems. In the present study, only the two broadband scales (internalizing and externalizing behavior problems) and the Total Behavioral Problems score were used. Reference values are provided by a healthy community sample of 2856 German children and adolescents between the ages of 4-18 years (Döpfner et al., 1998). This widely used measure has been shown to have excellent reliability and validity (Achenbach, 1991).

Socioeconomic status:

Socioeconomic status (SES) was calculated on the basis of both paternal occupation and maternal education on a 6-point scale. SES scores, ranging from 2-12 points, were classified into three social classes as follows: 2-5, low social class; 6-8, middle social class; and 9-12, upper social class. This measure has been proved to be a reliable and valid indicator of SES in our community (Landolt, Nuoffer, Steinmann, & Superti-Furga, 2002).

Pediatric Penile Perception Scale:

Patient's penile self-perception and quality of surgical result were assessed with the Pediatric Penile Perception Scale (PPPS), an instrument developed specifically for this study based on the work of Mureau, Slijper, Slob, & Verhulst (1995). The PPPS consists of 4 items that refer to important aspects of the surgical outcome: position and shape of meatus, shape of glans, shape of penile skin and general cosmetic appearance. Both patients and pediatric urologists

express their satisfaction with each penile aspect by means of a four-point scale that ranges from *very dissatisfied* (0) to *very satisfied* (3). By adding the raw scores of the 4 items, a PPPS-Total score (0-12) can be calculated. In the present study, six urologists of whom four were not affiliated to the Children's University Hospital Zurich were asked to rate the standardized photographs of the patients' penises (taken during the urological examination) by means of the PPPS. Afterwards, the PPPS scores of the six urologists were averaged in order to obtain an objective assessment of the quality of surgical outcome. Both interrater reliability (Intraclass correlation coefficient=.87) and internal consistency (Cronbach's α=.81-.84) of the PPPS were satisfactory. We will present the associations between PPPS scores (of both patients and urologists) and outcome measures in the present article. Results on differences between PPPS scores of patients, control subjects and urologists as well as the psychometric evaluation of the PPPS are to be reported elsewhere (Weber, Schönbucher, Gobet, & Landolt, 2007).

Medical factors:

Medical variables were taken from the patients' records. The severity of hypospadias was classified according to the intraoperative location of the meatus (glanular to perineal) as mentioned in the operation notes by the surgeon.

Psychosocial factors related to hypospadias:

Hypospadias patients were asked if they were ashamed of the appearance of their penis and if they had ever been teased about their penis. Both items were answered on a three-point scale (0=*not true*, 1=*somewhat true*, 2=*true*). The patients' mothers were asked whether or not they were concerned about the following three issues: their son's fertility, his sexual function and his future chances to find a partner. A sum score was computed by counting the number of maternal concerns, ranging from 0-3 points. Moreover, patients' mothers were asked to express on a 5-point scale how strongly they felt psychologically distressed by their son's hypospadias (*not at all*=0 to *very strongly*=4).

3.2.4 Statistical Analyses

Data were analyzed using the statistical package SPSS for Windows, release 14.0. All analyses were performed with two-sided tests. A value of $p \leq .05$ was considered significant. Chiefly, non-parametric statistical techniques were used (χ^2-tests, Mann-Whitney U-tests, Spearman-Brown rank correlations) because most variables showed either non-normal distribution or were categorical. In addition, effect sizes of group differences (Cohen's d) were

computed for the TACQOL- and CBCL-scales. Three linear regression models were set up using the CBCL Total Behavioral Problems score and the TACQOL-CF/-PF Total scores as dependents, which were normally distributed according to the Kolmogorov-Smirnov Test. The independent variables were chosen on the basis of previous knowledge and their statistical importance in the bivariate analyses.

3.3 Results

3.3.1 Health-Related Quality of life

Apart from positive emotional functions, boys with hypospadias showed significantly lower scores than the controls on all TACQOL-CF scales (table 4). Likewise, effect sizes were medium to large for all mean differences, except for positive emotions. The largest group differences were found with regard to the following items: Physical functions: nauseous (d=0.69), dozy (d=0.52); motor functions: playing (d=0.34), doing things handily/quickly (d=0.38); autonomy: sports/going out to play on my own (d=0.55), doing hobbies on my own (d=0.42); cognitive functions: concentrating (d=0.62), understanding schoolwork (d=0.56); social functions: other children asked me to play with them (d=0.46), impatient with parents (0.46), defiant with parents (d=0.63); negative emotional functions: sad (d=0.53), angry (d=0.53). In contrast, there were no significant differences between the groups on the mother-rated scales. Accordingly, effect sizes were small. A comparison between the hypospadias patients' self-rating and the mothers' rating showed that mothers rated their children's HRQoL significantly better than the children themselves on most TACQOL scales (table 4). Similarly, the mothers of the controls estimated their children's motor, social and negative emotional functions significantly better than their children themselves. However, the differences were clearly less pronounced than in the hypospadias group.

3.3.2 Psychological Adjustment

Mann-Whitney U-Tests displayed no significant group differences regarding psychological adjustment (table 4). Similarly, effect sizes of mean differences were small. Likewise, T-scores of hypospadias patients were highly comparable to those of the general population standardization sample (T=50) (Total Score: M=51.7, t=1.7, p=.10; Internalizing score: M=51.4, t=1.2, p=.24, Externalizing score: M=50.4, t=0.4, p=.72). Analysis of T-scores showed also that 6 (8.7%) participants with hypospadias and 10 (14.5%) control subjects had externalizing behavioral problems in the borderline/clinical range (χ^2=.32, p=.57), while 14

(20.3%) patients and 12 (17.4%) control subjects had internalizing behavioral problems in the borderline/clinical range (χ^2=.19, p=.66).

Table 4: Children's quality of life (TACQOL scores) and psychological adjustment (CBCL-raw-scores): Comparison with controls

	Boys operated for hypospadias		Boys operated for inguinal hernia		Effect size	
	Mean	SD	Mean	SD	d	p†
TACQOL-CF (n=136)‡						
Physical functions (0-32)	25.31***	5.16	27.79	3.15	.58	.005
Motor functions (0-32)	28.94***	2.77	30.25*	2.11	.53	.002
Autonomy (0-32)	30.53***	1.79	31.54	.95	.75	<.0001
Cognitive functions (0-32)	26.22*	4.11	29.12	2.97	.81	<.0001
Social functions (0-32)	25.68***	4.21	28.72*	2.82	.85	<.0001
Positive emotions (0-16)	12.66	2.20	13.32	1.86	.32	.09
Negative emotions(0-16)	9.69***	2.87	11.20*	2.85	.53	.003
TACQOL-PF (n=138)‡						
Physical functions (0-32)	27.44	5.31	28.60	3.14	.27	.55
Motor functions (0-32)	30.62	2.43	30.49	2.91	.05	.96
Autonomy (0-32)	31.23	1.80	31.52	1.34	.18	.36
Cognitive functions (0-32)	27.18	5.77	28.89	4.12	.34	.16
Social functions (0-32)	29.67	2.66	29.89	2.25	.09	.79
Positive emotions (0-16)	12.76	2.41	13.20	2.15	.19	.33
Negative emotions (0-16)	11.72	2.66	12.30	2.55	.22	.30
CBCL scales (n=138)‡						
Total score (0-236)	18.47	15.02	15.86	12.81	.19	.38
Internalizing score (0-62)	4.29	4.49	4.08	3.61	.05	.72
Externalizing score (0-66)	6.17	5.58	5.85	5.93	.06	.37

Note: Effect sizes according to Cohen: 0.20 small effect size, 0.50 medium effect size, >0.80 large effect size;
†U-Tests according to Mann-Whitney; ‡Higher scores indicate better HRQoL or more behavior problems, respectively
*p ≤ .05; **p ≤ .01; ***p ≤ .001: Significant differences between children's and parents' rating (U-Tests according to Mann-Whitney)

3.3.3 Determinants of HRQoL and Psychological Adjustment

Table 5 displays Spearman partial correlations controlling for subjects' age between sociodemographic, medical and psychosocial characteristics and TACQOL and CBCL scores, respectively. We also conducted the partial correlation coefficients controlling for severity of hypospadias, but did not find any marked differences to the results in table 5 (results available on request). Age showed several significant positive associations with self-reported HRQoL and significant negative associations with the CBCL Externalizing and Total score. Socioeconomic status was positively related to self- and mother-reported cognitive functions as well as with mother-reported autonomy. Furthermore, nationality was associated with mother-reported emotional functions, with Swiss children showing a higher degree of positive emo-

tions. Medical characteristics showed scarcely any significant correlations with outcome measures.

There were also a few significant correlations between variables related to the patient's experience of hypospadias (PPPS, being ashamed of penis, having been teased about penis) and HRQoL and psychological adjustment, respectively. Since the patients' PPPS was negatively associated with current age (r=-.32; p=.01), we conducted the correlations between the PPPS and the outcome measures separately for the age group below the median age (<9.97 years) and the one above the median age (>9.97 years). We found significant positive associations between the patients' PPPS and both mother-reported social functions (r=.57; p=0.00) and the TACQOL-PF Total score (r=.38; p=.05) in the older group, but did not find any significant correlations in the younger group. The correlations with regard to self-reported HRQoL and psychological adjustment did not markedly differ from the correlations calculated for the whole sample.

Finally, the mother's reporting of specific hypospadias-related concerns (fertility, sexual function, partner search) was negatively associated with self-reported autonomy (r=-.30, p=.03) and mother-reported positive emotional functions (r=-.27; p=.03). However, no significant correlation (r=-.25; p=.06 to r=. 18; p=.19) was found with respect to the extent to which mothers generally felt psychologically distressed by their sons' hypospadias ("How strongly do you feel psychologically distressed by your son's hypospadias?").

Table 5: Spearman partial correlation coefficients between TACQOL and CBCL scores and sociodemographic, medical and psychosocial variables (control variable: current age)

n=60-69	Current† age	SES	Natio-nality	Seve-rity	No. of surge-ries	Age at 1st operation	Days in hospital	Follow-up since last surgery	PPPS Uro-logists	PPPS Patient	Ashamed of penis	Being teased
TACQOL-CF												
Physical functions	.34**	.12	-.01	.11	-.14	-.07	-.08	.18	-.04	.07	-.17	-.16
Motor functions	.16	.16	-.14	-.16	-.02	.01	-.15	-.13	.21	.16	-.24	-.02
Autonomy	.38**	-.05	.13	-.01	.03	.15	.04	-.10	.01	.06	-.01	.05
Cognitive functions	.13	.31*	-.16	.06	-.15	.15	-.19	.22	.03	-.06	-.28*	.02
Social functions	.35**	.08	-.08	.08	-.14	-.07	-.15	.16	-.08	-.10	-.11	-.19
Positive emotions	.20	-.03	.12	.11	-.03	-.03	-.00	.03	.06	.15	-.06	-.08
Negative emotions	.27*	.00	.01	.16	.02	-.01	.15	.01	.00	-.07	-.30*	-.17
Total score	.44**	.18	-.06	.11	-.16	-.04	.15	.15	.07	-.00	-.26*	-.16
TACQOL-PF												
Physical functions	-.01	.12	.12	.16	.21	.19	.21	-.17	.11	.12	.11	-.15
Motor functions	.23	.07	.04	.22	.11	.04	.10	.05	.04	.23	.18	-.10
Autonomy	.29*	.33**	-.16	.24*	.04	-.15	.06	.17	.15	.11	-.21	-.11
Cognitive functions	.18	.29*	-.06	.23	-.08	-.18	.04	.21	.01	.18	-.16	.12
Social functions	.09	.04	.13	-.03	-.11	.03	-.02	.09	.07	.39**	-.17	-.08
Positive emotions	-.14	.03	-.30*	-.06	-.10	-.06	-.15	.15	-.13	-.02	-.23	.08
Negative emotions	.01	-.07	-.10	.00	-.25*	.10	-.13	.17	-.08	-.05	-.23	-.30*
Total score	.10	.19	-.08	.17	-.06	.01	.04	.15	-.02	.24	-.12	-.10
CBCL												
Total score	-.39**	-.02	.20	-.15	-.03	.01	-.08	.03	.16	.02	.10	.18
Internalizing score	-.23	.01	.13	-.16	-.11	-.04	-.08	-.05	.05	.02	.12	.35**
Externalizing score	-.41**	.06	.17	-.01	.05	.01	.04	.04	.22	-.06	.09	.05

Note: SES=socioeconomic status; nationality: Switzerland is coded 0, foreign countries are coded 1; † unadjusted spearman rank correlation coefficient
* $p \leq .05$. ** $p \leq .01$

Table 6: Summary of multiple regression analyses for predictors of children's quality of life (TACQOL Total scores) and psychological adjustment (CBCL Total score)

n=57-65	Equation 1: TACQOL-CF				Equation 2: TACQOL-PF				Equation 3: CBCL		
Variable	B	SEB	β	Variable	B	SEB	β	Variable	*B*	*SEB*	β
Age	1.87	.54	.40***	Age	.23	.57	.05	Age	-1.44	.60	-.30*
Severity	2.91	1.54	.24	Severity	3.56	1.38	.32*	Severity	-3.92	1.78	-.32*
Days in hospital	-.24	.15	-.21	PPPS Patient	2.65	.86	.38**	Days in hospital	.15	.16	.14
Ashamed of penis	-5.39	2.45	-.26*	Being teased	-8.90	2.64	-.40***	Being teased	6.85	3.21	.26*
								Nationality	8.59	3.84	.27*

Note: nationality: Switzerland is coded 0, foreign countries are coded 1

Equation 1: F=6.17; p<.0001; R^2=.29; R^2adj.=.24
Equation 2: F=6.03; p<.0001; R^2=.33; R^2adj.=.27
Equation 3: F=4.02; p=.004; R =.28; R^2adj.=.21
*$p \leq .05$. **$p \leq .01$. ***$p \leq .001$.

Table 6 shows the multiple regression equations with the two TACQOL Total scores and the CBCL Total score as dependent variables. With regard to the TACQOL-CF Total score, higher age and "being less ashamed of penile appearance" significantly contributed to the prediction of better HRQoL. In contrast, better mother-reported HRQoL was best predicted by the patient's experience of not having been teased about his penis, followed by a higher patient's PPPS and more severe hypospadias. In the CBCL regression model, however, more severe hypospadias was the strongest predictor for better behavioral adjustment, followed by higher age, Swiss nationality and having been teased less often. Overall, the selected variables accounted for 29% (R^2adj.=.24) (TACQOL-CF), 33% (R^2adj.=.27) (TACQOL-PF) and 28% (R^2adj.=.21) (CBCL) of the variance of the dependent outcome measures.

3.4 Discussion

This is the first comprehensive cross-sectional analysis on both HRQoL and psychological adjustment in children and adolescents with hypospadias. Consistent with our hypothesis, our results indicate that self-reported HRQoL of hypospadias patients might be diminished compared to healthy children. Unfortunately, no definition of clinically significant differences yet exists for the TACQOL measure. However, referring to Cohen (1988), we would consider effect sizes greater than 0.5 (medium effect size) to be clinically significant, which was the case for most dimensions.

Conversely, mothers evaluated their children's HRQoL more optimistically and similarly to the mothers of the control group. This divergent rating between children and mothers deserves attention. In a previous study with healthy Dutch children, parents rated their HRQoL also more optimistically than their children (Theunissen et al., 1998). However, mean differences between the ratings were less pronounced and presented only in some domains, similar to the findings in our control group. In contrast to our findings, previous HRQoL-studies among children with chronic diseases reported either a more pessimistically paternal view (Rüth, Landolt, Neuhaus, & Kemper, 2004) or similar scores between children and parents (Graf, Landolt, Capone Mori, & Boltshauser, 2006). Therefore, one may speculate that mothers of children with hypospadias might psychologically suppress the impact of hypospadias on their child's well-being. This assumption is supported by an observation we have made during the recruitment process. We asked non-participating families on the phone why they refused to participate in our study. 9 parents of the 25 non-participating families of the hypospadias group answered that they were not willing to deal with their sons' hypospadias (e.g. "We do

not want to talk about our son's hypospadias. This has never been an issue"). Another nine families, who had been debating the idea of participating for a considerably length of time, confirmed their participation after several telephone contacts, but kept neither the interview appointment nor returned the questionnaire. We have, however, not made similar observations neither with regard to the control group nor in previous studies on pediatric patients. Moreover, reports of adults show that most parents suppressed their son's hypospadias and kept it secret (Walker, 1998). Unfortunately, due to the lack of previous studies on HRQoL in this group of patients, we are unable to draw comparisons between our and previous findings. Thus, it is essential to conduct further studies in order to investigate whether the observed discrepancies between child- and mother-reported HRQoL can be confirmed in other samples. Should this be the case, it would be important to examine whether maternal defense mechanisms actually might account for the observed discrepancies, e.g. through qualitative interviews in which both patients and mothers would be asked about their experience of hypospadias.

Concerning behavioral adjustment, no group differences were found, which corresponds to findings of several recent studies (Mondaini et al., 2002; Mureau et al., 1997; Sandberg et al., 2001). This favorable outcome provides further empirical evidence that, nowadays, psychological adjustment of boys with hypospadias can be normal and might reflect the lately achieved improvements in medical treatment (e.g. in surgical techniques).

The identification of predictors of outcome revealed that younger age might be a significant risk factor for both an impaired child-reported HRQoL and behavioral maladjustment. These observations are surprising since boys with hypospadias are assumed to suffer mainly during puberty due to a changing physical self-perception as well as the increasing importance of sexuality (Bracka, 1999). The role of the patient's age in relation to his psychological well-being needs thus to be clarified in future studies.

A further astonishing finding is that more severe hypospadias predicted both better mother-reported HRQoL and psychological adjustment, which does not coincide with those from previous research (Mondaini et al., 2002; Mureau et al., 1997; Purschke & Standke, 1993; Sandberg et al., 1989, 2001). Since severe forms of hypospadias are associated with a more atypical appearance of the penis and higher complication rates[2], it is debatable that patients with more severe hypospadias are less able to deny their penile condition and thus develop

better coping strategies. On the other hand, it is also likely that children with more severe hypospadias get more medical attendance or more psychosocial support from their families.

No other medical factor turned out to be a risk factor for the patients' HRQoL and psychological adjustment, which corresponds with previous findings (Eray et al., 2005; Mondaini et al., 2002; Mureau et al., 1997; Sandberg et al., 2001). Instead, psychological factors, such as the patient's penile self-perception, his sense of shame and his experience of being teased about his penis by peers were important predictors of outcome. These results confirm our hypothesis and are in line with previous findings (Mureau et al., 1997). In addition, it is important to note that correlations calculated separately for age-groups showed that the significance of a positive penile self-perception for HRQoL may be higher in older than in younger children.

A further finding was that Swiss nationality predicted better psychological adjustment. Considering that foreign children usually live under more difficult conditions (e.g. less social acceptance) than Swiss children, the result is consistent with our hypothesis. However, one may speculate that the association may be caused by cultural differences in coping with hypospadias. Ultimately, a final result needs to be discussed: Maternal concerns about the consequences of hypospadias regarding their sons' sexuality were inversely related with some domains of HRQoL. Since such concerns are generally inappropriate, these findings point out the importance of extensive information for the parents about the implications of hypospadias.

The strengths of the present study include the use of multiple sources of information, the multidimensional and highly standardized assessment of HRQoL and psychological adjustment and the use of multivariate statistics. Furthermore, the study involved a control group equally large to the patient sample and successfully matched regarding current age and age at first operation. Nevertheless, some limitations need to be mentioned. Firstly, since the present study bears an exploratory character, we presented a high number of bivariate correlations, which increased the probability of alpha error. Yet, we countered this problem by conducting multivariate analyses. Secondly, the markedly higher response rate for the hypospadias patients (75.5%) than for the comparison males (58.8%) might have biased the results. On the one hand, non-participating hypospadias patients may be particularly ashamed of their condition and the level of HRQoL and psychological adjustment of the participating patients may

therefore be overestimated in the present study. On the other hand, the control group is more self-selected for volunteerism due to their lower participation rate. Since research has demonstrated that volunteers are generally better adapted than those in the general population (Kazdin, 2003), outcome scores of the control subjects might be more likely to be an overestimation than the ones of hypospadias patients. Finally, the TACQOL and the CBCL are generic measures of HRQoL and psychological adjustment, respectively, and may thus lack sensitivity for specific problems of children and adolescents with hypospadias (e. g. micturition problems, sexual anxieties). To our knowledge there are no hypospadias-specific measures for HRQoL and psychological adjustment.

The present study provides some issues for future research. Since it is the first investigation of HRQoL of children and adolescents with hypospadias, our study may be considered to be a basis for further studies conducted in other countries and with samples that involve younger children. In particular, prospective studies are necessary to fully understand the factors that may influence long-term outcome. Despite the statistical significance of our findings, multivariate analyses explained only between 28 % and 35 % of the variability in HRQoL and psychological adjustment, respectively. Thus, variables not included in this study, such as the patient's coping strategies, may be of particular importance. Furthermore, the development of a hypospadias-specific HRQoL measure should be taken into consideration. Ultimately, it would be interesting to compare our results with data from untreated children with hypospadias since genital surgery is assumed to psychologically traumatize the patients (Bracka, 1999; Mureau et al., 1997). This issue should be raised since in mild hypospadias surgery is exclusively performed due to cosmetic reasons (Duckett, 1998). Therefore, one might speculate that untreated patients would show better psychological and psychosexual adjustment.

Finally, our findings implicate some consequences for clinical management. To guarantee a comprehensive treatment for children with hypospadias, not only surgical results should be carefully evaluated, but also the childrens' HRQoL. In doing so, both children and parents should be asked, since the results showed that proxy- and self-perception of HRQoL can considerably diverge. Surgeons need to be aware that HRQoL of children with hypospadias might be impaired by negative penile self-perception, sense of shame or teasing remarks from peers. As previous studies have provided evidence that social skill training can improve social functioning and quality of life in pediatric patients (Barakat et al., 2003; Grey, Boland, Davidson,

& Tamborlane, 2000; Varni, Katz, Colegrove, & Dolgin, 1993), one may argue that psychological support for the development of sufficient self-confidence may help hypospadias patients to accept their penile appearance as well as to encourage them to defend themselves against teasing remarks from their peers.

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4 Psychosexual Development of Children and Adolescents with Hypospadias

4.1 Abstract

Aim: A comprehensive, cross-sectional investigation of the psychosexual development of boys operated on for hypospadias in comparison to a healthy control group.

Methods: 68 children and adolescents (7-17 years) operated on for hypospadias were examined by means of a standardized interview assessing penile self-perception, gender-role behavior, sexual experiences and sexual attitude. Scores were compared to an age-matched control group consisting of 68 boys after hernia repair. Predictive values of medical variables as well as the patients´ knowledge of hypospadias were assessed.

Main outcome measures: The Pediatric Penile Perception Score, the Gender-role Questionnaire by Ijntema and Cohen-Kettenis and a self-developed questionnaire on first sexual experiences and sexual attitude comprised the standardized assessment instruments.

Results: Boys with hypospadias did not significantly differ from the control subjects with regard to penile self-perception, gender-role behavior, first sexual experiences and sexual attitude. Younger age and the patient's knowledge of hypospadias predicted a more positive penile self-perception, while a more pronounced masculine gender-role behavior was best predicted by younger age at final surgery.

Conclusions: Boys with corrected hypospadias may show a psychosexual development that is similar to healthy children. Puberty could be a critical time for the patients however, during which they might require regular urological follow-ups and may benefit from age-appropriate information about their penile condition. Moreover, the later corrective surgery is completed, the more likely the patients may become insecure with regard to gender-role behavior.

4.2 Introduction

Hypospadias is the most common malformation of the penis with a reported incidence ranging up to 3 per 1000 male births (Dolk, 2004). It is manifested by an abnormal position of the urethral opening, which lies proximal to its normal location in the glans, on the ventral surface of the penis or in the perineum. In most hypospadias, the prepuce is split and severe forms are associated with a curvature of the penis (chordee) (Duckett, 1998).

The goal of hypospadias surgery is to achieve a normal looking penis, allow unimpaired micturition and enable full sexual function (Duckett, 1998). Since it is believed that hypospadias should be surgically corrected prior to the patients' awareness of his penile malformation, most surgeons recommend surgery as early as the first 12 months to avoid any negative impact on the patients' psychosexual development (APA, 1997; Bracka, 1999; Manzoni, Bracka, Palminteri, & Marocco, 2004; Schultz, Klykylo, & Wacksman, 1983). These suggestions on the timing of surgical correction, however, are not evidence-based.

The term *psychosexual development* implies that sexual development is not only a physical process but also interacts with psychological factors. It comprises many components such as the development of gender identity, gender-role behavior, body image and sexual behavior habits (Schuhrke, 1997). Several factors related to hypospadias are assumed to expose the patients to an increased risk of psychosexual maladjustment. Firstly, the patients generally experience the stressors of genital surgery (Mureau, Slijper, Slob, & Verhulst, 1997; Sandberg et al., 2001). Secondly, hypospadias surgery does not result in a perfectly normal penile appearance and can entail some functional complications (e.g. fistulas, stenoses) (Duckett, 1998). Ultimately, there is increasing evidence that prenatal exposure to androgens not only induces the sexual differentiation of the human genitalia but also influences postnatal sex-typed behavior patterns by engendering sex-specific brain structures. Most of the evidence derives from studies on individuals with DSD, who are exposed to non physiological androgen levels (Cohen-Bendahan, van de Beck, & Berenbaum, 2005). Because the faulty embryogenesis of the penile urethra in hypospadias is assumed to be related to abnormalities in prenatal androgen exposure and/or androgen receptor defects (Baskin, 2004), Sandberg et al. (1995) hypothesized that children born with hypospadias might show a more atypical gender-role behavior as well. This hypothesis has not been confirmed to date.

However, despite the common assumption that hypospadias may affect the children's psychosexual development, only few studies report on the patients' psychosexual adjustment during childhood and adolescence (Schönbucher, Weber, & Landolt, in press). Previous findings indicate that boys with hypospadias suffer more often from negative genital appraisal and sexual inhibitions than healthy age-matched controls, but do not differ from healthy adolescents with regard to sexual activity, number of coitus partners and sexual desire (Mondaini et al., 2002; Mureau et al., 1995a, 1995b). Furthermore, the studies show that psychosexual development is partly affected by the severity of hypospadias and the number of hospitalizations, but not by the quality of surgical outcome, age at final surgery, number of operations and type of surgical procedure (Mondaini et al., 2002; Mureau et al., 1995a, 1995b, 1996, Sandberg et al., 1989, 1995; Schönbucher et al., in press). Results are inconsistent with regard to gender-role behavior and age at first sexual experiences (Mondaini et al., 2002; Mureau et al., 1995a; Sandberg et al., 1989, 1995; Schönbucher et al., in press).

4.3 Aims

The present study aimed at a comprehensive, cross-sectional investigation of the psychosexual development of boys operated on for hypospadias in comparison to a healthy control group. With reference to previous results and theoretical assumptions, we considered the patients' penile self-perception, gender-role behavior, first sexual experiences and overall sexual attitude as the most relevant constructs for our study. We hypothesized that boys with hypospadias would show a more negative penile appraisal, a less pronounced masculine gender-role behavior, a delay in first sexual experiences and a more negative sexual attitude than the control subjects. In addition, we postulated that severity of hypospadias, number of operations, length of hospitalization, a higher age at operation, a shorter follow-up and a less favorable surgical result bear a negative influence on the psychosexual outcome. Moreover, we expected the patients who were informed about their hypospadias to show a more positive psychosexual development than patients who were not aware of their hypospadias.

4.4 Methods

4.4.1 Procedure

The present study was approved by the local research ethics committee. Children and parents were informed about the study by letter and were requested to return written informed consent. The participating children and adolescents were assessed by means of a standardized interview that was usually conducted at the hospital. In addition, hypospadias patients were

offered a urological examination, during which standardized photographs of their genitalia were taken.

4.4.2 Subjects

Patients:

Children and adolescents operated on for hypospadias at the University Children's Hospital Zurich (1991-2005) were eligible for this study (n=147). Inclusion criteria for participation in the study were (a) age between 6 and 17 years, (b) command of the German language, (c) follow-up since last surgery ≥1 year, (d) no current postoperative treatment of hypospadias, and (e) residence in Switzerland. Children with chronic diseases, disabilities or hypospadias as a concomitant phenomena in a defined DSD were excluded.

102 children fulfilled the recruitment criteria, of which 68 agreed to participate (66.6%). Demographic and medical characteristics of the sample are listed in table 7. No significant differences between participants and non-participants with regard to the child's age (U=789.5, p=.60), socioeconomic status (U=480.0, p=.45), nationality (χ^2=.20, p=.79) and medical characteristics such as severity of hypospadias (χ^2=.69, p=.35), age at first operation (U=796.5, p=.64), age at final operation (U=725.5, p=.69), number of operations (U=831.0, p=.85), days in hospital (745.5, p=.60), and follow up since last surgery (U=621.5, p=.17) were observed. Of the 68 participating patients, 65 could be interviewed about penile self-perception (in 3 cases, the mother did not allow the interviewer to refer to hypospadias), 47 about gender-role behavior (because the measure applied is only suitable for children up to 12 years) and 20 about both sexual milestones and sexual attitude. The latter consisted of children who were at least 12 years old and who appeared to be sufficiently mature to be asked about sexual matters.

Control group:

Each participant of the hypospadias group was matched with regard to both current age and age at first operation with a boy who was treated for an inguinal hernia. In order to have an equally large control group, 116 children were contacted of which 48 (39.3%) refused to participate. Control subjects did not significantly differ from the hypospadias group with regard to nationality, socioeconomic status, age at assessment and age at first operation (table 7). However, the boys with hypospadias had their final surgery at a higher age, had more operations, were hospitalized for a longer period and showed a shorter follow-up since their last

surgery. All 68 control subjects could be asked about penile self-perception, 49 about gender-role behavior and 25 about sexual milestones and sexual attitude.

Table 7: Demographic and medical characteristics of patients and controls

	Patients (n=68)	Controls (n=68)	p†
Age			
Mean (SD)	10.81 (3.18)	11.05 (3.18)	.61
Median (Range)	9.84 (6.66-17.34)	10.39 (6.80-18.23)	
Nationality (%)			
Swiss	58.5	69.2	.27
Foreigners*	41.5	30.8	
Socioeconomic status (%)			
Lower	25.0	17.2	.47‡
Middle	45.3	56.2	
Upper	29.7	26.6	
Age at 1st operation			
Mean (SD)	3.18 (2.45)	2.97 (2.87)	.21
Median (Range)	2.30 (0.20-11.87)	2.02 (0.20-14.18)	
Age at final operation			
Mean (SD)	4.26 (3.18)	3.09 (2.96)	.008
Median (Range)	2.92 (0.78-12.29)	2.05 (.20-14.18)	
No. of operations			
Mean (SD)	1.91 (2.18)	1.10 (0.31)	<.0001
Median (Range)	1.00 (1.00-15.00)	1.00 (1.00-2.00)	
Days in hospital			
Mean (SD)	11.87 (13.27)	2.63 (1.18)	<.0001
Median (Range)	8.00 (1.00-77.00)	2.00 (1.00-7.00)	
Follow-up since last surgery			
Mean (SD)	6.32 (3.23)	7.96 (3.52)	.002
Median (Range)	6.28 (1.00-15.15)	7.46 (0.80-16.13)	
Severity of hypospadias (%)			
glanular	19.1	-	-
coronal	35.3	-	-
subcoronal	8.8	-	-
penil	32.4	-	-
penoscrotal	4.4	-	-

Note: †U-Tests according to Mann-Whitney for dichotomous variables, χ^2-Tests for categorical variables; ‡U-Test according to Mann-Whitney based on SES scores; *mainly originating from former Yugoslavia

4.4.3 Main Outcome Measures

Pediatric Penile Perception Scale:

Patient's penile self-perception and quality of surgical result were assessed with the Pediatric Penile Perception Scale (PPPS), an instrument developed specifically for this study (Weber, Schönbucher, Gobet, & Landolt, 2007). The PPPS consists of 4 items that refer to important aspects of the surgical outcome: position and shape of meatus, shape of glans, shape of penile skin and general cosmetic appearance. Both patients and pediatric urologists express their

satisfaction with each penile aspect by means of a four-point scale that ranges from *very dissatisfied* (0) to *very satisfied* (3). By adding the raw scores of the 4 items, a PPPS-Total score (0-12) can be calculated. In the present study, six blinded urologists were asked to rate the standardized photographs of the patients' penises (taken during the urological examination) by means of the PPPS. Their scores were averaged to obtain an objective outcome measurement. Both interrater reliability (Intraclass correlation coefficient=.87) and internal consistency (Cronbach's α PPPS Patients=.81; PPPS Urologists=.84) of the PPPS were satisfactory. The development and psychometric evaluation of the PPPS are to be reported elsewhere (Weber et al., 2007).

Gender-role behavior:

The Gender-role Questionnaire by Ijntema and Cohen-Kettenis (Cohen-Kettenis & Pfäfflin, 2003) is a Dutch 32-item questionnaire designed for children up to 12 years with questions regarding hobbies, sports, games, friends, clothing, identity and self-attribution of gender characteristics. Children are asked to choose one out of three gender-typical items (e. g. *Which is your favorite sport? football/swimming/ballet*). By summing 31 of the 32 questions, a total score is calculated ranging from 31 to 93. Higher values indicate a more masculine behavior. The questionnaire shows large gender differences and has an excellent Cronbach's Alpha of .92 (Cohen-Kettenis & Pfäfflin, 2003). In the present study, we used an authorized German translation of the questionnaire, which was developed by Thyen, Kleinemeier and Fischer at the University Hospital Lubeck (Germany) according to international translation standards (Medical Outcomes Trust, 1997).

Sexual experiences:

Adolescents were asked whether or not and at what ages they have experienced different sexual milestones (see table 8). In addition, they were asked if they regularly masturbated and if they currently had a girlfriend/boyfriend.

Sexual attitude:

By means of a three-point scale (0=*not true*, 1=*somewhat true*, 2=*true*), adolescents were asked to express their level of agreement with the following four statements: Sexuality is 1) something enjoyable, 2) something important, 3) something embarrassing and 4) something I am not interested in. After recoding the negatively formulated items, a sum score was computed (0-8) showing a Cronbach's Alpha of .58. Higher values indicate a more positive sexual attitude.

Sociodemographic and medical characteristics:

Sociodemographic and medical variables were taken from the patients' records. Socioeconomic status (SES) was calculated on the basis of both paternal occupation and maternal education on a 6-point scale. SES scores, ranging from 2-12 points, were classified into three social classes as follows: 2-5, low social class; 6-8, middle social class; and 9-12, upper social class. This measure has been shown to be a reliable and valid indicator of SES in our community (Landolt, Nuoffer, Steinmann, & Superti-Furga, 2002).

Knowledge of hypospadias:

Patients were asked if they knew the reason for having been operated on their penis. If the reported reasons were related to hypospadias in some way (e.g. "I was not able to direct my urine stream properly"), the subjects were categorized as having some knowledge of their hypospadias (1). If the reported reasons were not related to hypospadias (e.g. "I was circumcised") or if the subjects were not able to name a reason, they were categorized as being not aware of their hypospadias (0).

4.4.4 Statistical Analyses

Data were analyzed using the statistical package SPSS for Windows, release 14.0. (SPSS Inc., Chicago, USA). All analyses were performed with two-sided tests. A value of $p \leq .05$ was considered significant. For categorical comparisons, χ^2-tests were used. In case that >20% of the cells showed an expected frequency <5, Cramer's V was applied. For group comparisons with regard to the PPPS and the sexual attitude score Mann-Whitney U-tests were performed because both scales showed non-normal distribution. For group comparisons regarding gender-role behavior and age at sexual milestones, t-tests were conducted because both variables were normally distributed according to the Kolmogorov-Smirnov test. In addition, effect sizes (ES) of mean differences according to Cohen were computed, using pooled standard deviations of the two groups. An ES of 0.2 is considered to be small, an ES of 0.5 to be moderate, and an ES of 0.8 to be large (Cohen, 1988). Spearman partial correlations controlling for subjects' age were computed between medical and sociodemographic variables and the PPPS score and gender-role behavior, respectively. Calculations of correlations regarding sexual experiences and sexual attitude were not considered to be adequate due to the small number of patients (n=20) interviewed. Moreover, two linear regression models were set up using the gender-role behavior score and the square root transformed PPPS score, which was normally distributed according to the Kolmogorov-Smirnov Test. The independent variables were cho-

sen on the basis of previous knowledge and their statistical importance in the bivariate analyses.

4.5 Results

4.5.1 Psychosexual Development

No significant group differences between hypospadias patients and the control group were found with regard to penile self-perception, gender-role behavior and sexual attitude (table 8). Accordingly, effect sizes were small. Overall, both groups showed high satisfaction with penile appearance and showed an overall positive sexual attitude. Likewise, adolescents with hypospadias and the control subjects did not differ with regard to their sexual experience, age at sexual milestones and current relationship status (table 8). However, adolescents with hypospadias reported significantly more often that they already had experienced orgasm through masturbation. Furthermore, regarding age at first petting, effect size was large suggesting that adolescents with hypospadias experienced their first petting at a higher age. Yet, due to the small number of adolescents who already had experienced petting, effect size did not reach significance (Cl 95%: -0.54 - 2.08).

4.5.2 Determinants of Penile Self-Perception and Gender-role Behavior

Table 9 displays correlations between sociodemographic and medical characteristics as well as knowledge of hypospadias with PPPS score and gender-role behavior, respectively. The PPPS score was significantly negatively associated with current age and positively related to knowledge of hypospadias. Gender-role behavior was significantly negatively correlated to the patients' age at last surgery and positively associated with follow-up since last surgery. There was also a correlation between gender-role behavior and age at first surgery, but the association scarcely reached significance (p=.07).

Table 8: Patients' PPPS scores, gender-role behavior, sexual attitude and sexual experiences: Comparison with controls

	Patients	Controls	d	P value[†]
PPPS score (n=133)				
M (SD)	9.51 (2.12)	9.77 (1.97)	-0.12	.51
Gender-role behavior (n=96)				
M (SD)	69.64 (4.84)	68.93 (3.79)	0.16	.27
Sexual attitude (n=45)				
M (SD)	6.65 (1.98)	6.02 (2.53)	0.28	.59
Sexual experiences (n=45)				
Ever been in love? (f, f%)				
yes	19 (95.0)	21 (84.0)		.24
no	1 (5.0)	4 (16.0)		
Age at being in love the first time				
M (SD)	10.47 (2.93)	10.19 (2.93)	0.10	.73
Ever been dating? (f, f%)				
yes	16 (84.21)	18 (85.7)		.89
no	3 (15.79)	3 (14.3)		
Age at first date				
M (SD)	12.00 (2.61)	11.72 (2.35)	0.11	.70
Ever had a girlfriend/boyfriend? (f, f%)				
yes	13 (65.0)	17 (68.0)		.83
no	7 (35.0)	8 (32.0)		
Currently a girlfriend/boyfriend? (f, f%)				
yes	4 (20.0)	5 (20.0)		1.00
no	16 (80.0)	20 (80.0)		
Ever masturbated? (f, f%)				
yes	15 (75.0)	15 (60.0)		.29
no	5 (25.0)	10 (40.0)		
Age at masturbating the first time				
M (SD)	12.67 (1.59)	12.27 (1.67)	0.25	.39
Do you masturbate regularly? (f, f%)				
yes	5 (33.3)	9 (60.0)		.14
no	10 (66.7)	6 (40.0)		
Ever had orgasm when masturbating? (f, f%)				
yes	15 (100.0)	11 (73.3)		.03
no	0 (0.0)	4 (26.7)		
Age at first orgasm				
M (SD)	13.36 (1.50)	13.45 (1.57)	-0.06	.89
Ever French kissed? (f, f%)				
yes	13 (65.0)	15 (60.0)		.73
no	7 (35.0)	10 (40.0)		
Age at first French kiss				
M (SD)	13.15 (2.00)	13.40 (0.83)	-0.16	.93
Ever fondled? (f, f%)				
yes	12 (60.0)	13 (52.0)		.59
no	8 (40.0)	12 (48.0)		

Table 8 continued

	Patients	Controls	d	P value†
Age at fondling the first time				
M (SD)	13.50 (1.83)	13.15 (1.28)	0.22	.54
Ever had petting? (f, f%)				
yes	4 (20.0)	6 (24.0)		.75
no	16 (80.0)	19 (76.0)		
Age at first petting				
M (SD)	14.50 (1.29)	13.67 (0.82)	0.77	.35
Ever had intercourse? (f, f%)				
yes	1 (5.0)	5 (20.0)		.14
no	19 (95.0)	20 (80.0)		
Age at first intercourse				
M (SD)	15.00 (-)	14.40 (0.55)	N/A	.67

Note: †U-Tests according to Mann-Whitney for non-normally distributed continuous variables, t-tests for normally distributed continuous variables, χ^2-tests or Cramer's V for categorical variables; PPPS Patient=patient's penile self-perception

Table 9: Spearman partial correlation coefficients between patients' PPPS scores and gender-role behavior and sociodemographic, medical and psychosocial variables (control variable: current age)

	PPPS Patient	Gender-role behavior
Current Age	-.33*	-.22
SES	.10	.17
Nationality	.02	-.20
Severity	-.02	.08
No. of operations	.12	-.10
Age at 1st operation	-.10	-.28
Age at final operation	.05	-.36*
Days in hospital	.11	-.13
Follow-up since last surgery	-.09	.38*
PPPS Urologists	.15	.03
Knowledge of hypospadias	.35**	.01

Note: nationality: Switzerland is coded 0, foreign countries are coded 1; PPPS Patient=patient's penile self-perception; PPPS Urologists=quality of surgical result; SES=socioeconomic status

$p \leq .05$. ** $p \leq .01$

Statistics for the multiple regression analyses are presented in table 10. With regard to the PPPS score, younger age and knowledge about hypospadias significantly contributed to the prediction of a more positive penile self-perception. In contrast, a more pronounced masculine gender-role behavior was best predicted by younger age at final surgery. Overall, the selected variables accounted for 27% and 26%, respectively, of the variance of the outcome measure.

Table 10: Summary of multiple regression analyses for predictors of patients' PPPS scores and gender-role behavior

	Equation 1: PPPS Patient			Equation 2: Gender-role behavior		
Variable	B	SEB	β	B	SEB	β
PPPS Urologist	.10	.14	.11	-.08	.40	-.03
Age at final operation	.03	.09	.06	-1.07	.36	-.50**
Current age	-.19	.09	-.31*	-.22	.58	-.07
Knowledge of hypospadias	1.35	.49	.37**	.98	1.66	.10

Note: PPPS Patient=patient's penile self-perception; PPPS Urologists=quality of surgical result
Equation 1: F=4.06; p<.01; R^2=.27; R^2adj.=.20
Equation 2: F=2.77; p<.05; R^2=.26; R^2adj.=.16
*$p \leq .05$. **$p \leq .01$. ***$p \leq .001$.

4.6 Discussion

The present study reports on a comprehensive, cross-sectional investigation of the psychosexual development of boys operated on for hypospadias in comparison to a healthy age-matched control group. As opposed to both our hypothesis and previous results (Mondaini et al., 2002; Mureau et al., 1995a, 1995b), boys with hypospadias showed an overall positive penile self-perception that was similar to those of the control subjects. This unexpected finding might be a result of the lately achieved improvements in treatment which result in better postoperative functional and cosmetical outcomes. Yet, it can be argued that the different age ranges of samples between the studies have contributed to the inconsistent results since previous studies included children and adolescents at somewhat higher mean ages (Mondaini et al., 2002; Mureau et al., 1995a, 1995b).

Likewise in contradiction to our hypotheses are the actual findings that boys with hypospadias did not differ from the control subjects with regard to gender-role behavior, first sexual experiences and sexual attitude. This corresponds with findings from previous studies by Sandberg et al. (1995) and Mureau et al. (1995a) and suggests that children and adolescents with hypospadias may show a psychosexual development that is similar to healthy children. However, we did not elicit any information from the subjects' experiences of their sexual milestones, e. g. to what degree they had suffered from sexual anxieties. The quality, rather than the quantity, of the patients' sexual activity may be more important for their well-being and should be examined in future studies.

This study also examined medical, sociodemographic and psychosocial correlates of penile self-perception and gender-role behavior. Both bivariate and multivariate analyses revealed

that higher age and not being aware of one's hypospadias were negatively associated with penile self-perception. The finding that adolescent patients are less satisfied with penile appearance than younger children is in line with previous results (Mureau et al., 1995b) and might be the consequence of a change in physical self-perception as well as an increasing importance of sexuality during puberty. The finding that patients who were informed about their hypospadias showed a more positive penile self-perception is also consistent with our hypothesis and reinforces the claim of self-help groups and experts in the field of DSD for making age-appropriate information available for patients about their penile condition (ISNA, 1994, 2006; Lee, Houk, Ahmed, & Hughes, 2006). Furthermore, it is interesting that medical factors such as the severity of hypospadias and the urologists' assessment of the quality of surgical result did not reveal to be important predictors of penile self-perception. This indicates that the latter is predominantly subjective and confirms the finding of Mureau et al. (1996) that the patients' and surgeons' penile appraisal can considerably diverge from each other.

As for masculine gender-role behavior, a younger age at final surgery was the strongest predictor. This result supports the presumption that early surgical treatment may prevent negative effects on the patients´ gender development. However, from our point of view, it is arguable whether an accumulation of feminine behavior patterns is indicative of psychosexual maladjustment considering that gender differences in behavior preferences have been decreasing since the issue of gender equality was raised. Moreover, in a previous study we found no association between age at surgical intervention, HRQoL and psychosocial adjustment (Schönbucher et al., in press). We consider both of these outcome variables to be more significant indicators of the patients' well-being than gender-role behavior. Yet, one may argue that the actual finding that follow-up since last surgery was positively correlated with masculine behavior patterns confirms the assumption that genital surgery may affect the patients' psychosexual development, at least for a short term.

Finally, one further issue needs to be discussed. Our findings fail to support the hypothesis that failures in prenatal androgen synthesis and/or androgen receptor defects influence postnatal sex-typed behavior patterns of hypospadias patients. Firstly, we did not find any group differences regarding gender-role behavior. And secondly, severity of hypospadias was not related to gender-role behavior, although more severe forms of hypospadias are supposed to

be a result of a more severe degree of endocrine dysfunction (Gearhart, Donohoue, Brown, Walsh, & Berkovitz, 1990; Sandberg et al., 1995; Shima et al., 1986).

4.6.1 Limitations

The strength of the present study lies in its significant contribution to the investigation of the psychosexual development of children and adolescents with hypospadias. Moreover, the study is based upon a precisely defined sample as well as a control group that is equally large and successfully matched regarding age and age at first operation. Yet, some limitations deserve closer attention. Firstly, since the present study bears an exploratory character, we presented a high number of bivariate correlations, which increased the probability of alpha error. We countered this problem by conducting multivariate analyses. Secondly, our sample is small, particularly with regard to the number of adolescents who were interviewed about their first sexual experiences and sexual attitude, which restricts the generizability of our results. Thirdly, despite an acceptable response rate of 66.6%, non participating hypospadias patients may be particulary ashamed of their condition and thus may have more sexual problems than participating patients. Finally, apart from the Gender-role Questionnaire by Ijntema and Cohen-Kettenis (Cohen-Kettenis & Pfäfflin, 2003), we applied self-developed measures. While the Penile Perception Scale proved to be a reliable measure, the Sexual Attitude Scale showed only a Cronbach's Alpha of .58. Unfortunately, we could not find any appropriate validated German measures of psychosexual development.

4.6.2 Conclusions

The aim of the present study was to shed light on the psychosexual development of boys with hypospadias. The study bears an exploratory character and our data suggest therefore several issues for future research activities. Our study may be considered to be a basis for further studies conducted with larger samples with different age ranges. Prospective studies are necessary to fully understand the factors that may influence long-term outcome. The role of the child's age at surgical correction requires further clarification in particular. Despite the statistical significance of our findings, multivariate analyses of penile self-perception and gender-role behavior explained only 27% and 26%, respectively, of the outcome. Thus, variables not included in this study, such as the patient's and parents' coping strategies, may be of particular importance. Finally, the development and validation of standardized measures of different constructs of psychosexual development should be taken into consideration. Ideally, they would not only provide reference data about the psychosexual development of healthy chil-

dren but also be sensitive to the specific challenges of hypospadias. To the best of our knowledge, there are excellent measures of psychosexual differentiation for both healthy children and children with DSD (Meyer-Bahlburg, Sandberg, Yager, Dolezal, & Ehrhardt, 1994). However, validated measures of other constructs of psychosexual development such as first sexual experiences, sexual behavior habits and sexual problems have yet to be developed.

Ultimately, our findings also implicate some issues for clinical management. Although our results suggest that boys with hypospadias may show a healthy psychosexual development, they also indicate that puberty might be a critical time for the patients, who therefore may benefit from regular urological follow-ups until young adulthood. Apart from urological examinations, the patients should be asked how they feel about sexual matters and they should be provided with age-appropriate information about their penile condition. Moreover, surgeons must be aware that the objective surgical outcome and severity of hypospadias might be secondary for the patients' psychosexual development during childhood. Yet, our results might suggest that the later corrective surgery is completed the more likely the patients may become insecure with regard to gender-role behavior.

4.7 References

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5 General Discussion

The objective of this Ph.D.-project was to undertake a comprehensive, cross-sectional investigation of the psychosexual development, HRQoL and psychological adjustment of children and adolescents operated on for hypospadias in comparison to a healthy age-matched control group. Furthermore, the study aimed to identify sociodemographic, medical and psychosocial predictors of outcome.

This final chapter begins with a summary and overall discussion of the results, followed by methodological considerations with regard to the study design. Conclusions are drawn and suggestions for future research activities and clinical practice are outlined.

5.1 Summary and discussion of the results

It was hypothesized that, compared to healthy boys, children and adolescents with surgically corrected hypospadias would show an impaired psychological adjustment, HRQoL and psychosexual development. Consistent with our hypotheses, the results indicate that self-reported HRQoL of hypospadias patients may be lower than HRQoL of healthy children. Yet, we did not find any group differences with regard to mother-reported HRQoL, psychological adjustment and psychosexual development such as penile self-perception, gender-role behavior, first sexual experiences and sexual attitude.

This positive outcome is in line with findings of several methodologically sound studies which were published in recent years (Mondaini et al., 2002; Mureau et al., 1995a, 1997; Sandberg et al., 1995, 2001) but contradict results of somewhat older and methodologically less sound studies (e.g. Purschke & Standke, 1993; Sandberg et al., 1989). It is debatable whether the more negative outcomes are the results of the studies' lower methodological quality (Schönbucher, Weber, & Landolt, in press). Main weaknesses of the latter include small sample size, lack of comparison data and the use of partly non-validated measures. Non-representative samples restrict the generalizability of results, non-validated measures are bound to produce non-valid results and only the inclusion of a control group allows an objective interpretation of results. Therefore, the methodological shortcomings of older studies might have biased the results. Although it is not clear how these shortcomings have influenced the results, they might have contributed to an underestimation of the children's psychological and psychosexual adjustment. On the other hand, it can be argued that surgical treatment strategies have advanced over the last decades: Patients are operated at younger ages

and both surgical techniques and anesthesia have been improved. Furthermore nursing practice has adapted to children's needs as for example by the rooming-in of parents. These improvements in treatment are presumed to have made hypospadias repair to be less stressful for the children than it used to be: "There is less physical trauma, with fewer complications, and better postoperative function and cosmesis. Hospitalizations are fewer and shorter,…We anticipate that patients will grow up to have fewer psychological problems and negative feelings [regarding sexual matters] than their predecessors" (Bracka, 1999). However, the fact that we did not find any associations between the outcome and quality of surgical result, length of hospitalization and number of operations does not confirm Bracka's thesis. To date the association between progress in medical treatment of pediatric patients and improvements in HRQoL and psychosocial adjustment, respectively, has not been empirically proved. Yet, it can be assumed that such an association exists.

The finding that boys with hypospadias showed an overall positive penile self-perception which did not differ from that of the control subjects, however, was surprising and contradicts previous results (Mondaini et al., 2002; Mureau, Slijper, Slob, & Verhulst, 1995b). This unexpected result might again reflect the lately achieved improvements in postoperative functional and cosmetic outcomes. But it can also be argued that the different age ranges of samples between the studies have contributed to the inconsistent results since previous studies included children with higher mean ages. It is assumable that older children who have already reached puberty tend to negatively appraise their genitals more often. This assumption is supported by our finding that penile self-perception was negatively associated with current age.
Further, another explanation for our results contradicting previous findings could be the different operationalization of penile self-perception. Whereas the PPPS consists of four items assessing satisfaction with position of meatus, shape of glans, penile skin and general appearance, Mureau's (1995b) Genital Perception Scale comprises eight items of which three were related to penile size. Penile size is considered to be a main concern of hypospadias patients since an underdevelopment of the penile size is more often encountered among hypospadias patients than among healthy boys (Duckett, 1998). As the PPPS does not contain an item related to penile size (because penile size is not amenable for surgery and thus not an indicator for the quality of surgical result) one could argue that penile self-perception of the patients might be overestimated in the present study. However, in Mureau's study (1995b) children with hypospadias showed a negative genital appraisal on all items except for quality of erec-

tion. This findings contradict the assumption that satisfaction with penile size is a more important determinant of overall penile satisfaction than other penile aspects (e. g. position of meatus). Mondaini et al. (2002) have not reported how genital appraisal was assessed in their study. Thus, we are not able to compare our operationalization of penile self-perception with the one of Mondaini et al.

In contrast to the favorable outcome regarding psychological adjustment and psychosexual development, most dimensions of self-reported HRQoL of hypospadias patients were diminished when compared to the control subjects. Although a cut-off score for clinically significant group differences does not yet exist for the TACQOL measure, we consider the observed group differences to be clinically significant since effect sizes were mainly medium (0.5) to large (0.8). These findings are striking and suggest that hypospadias and its treatment may affect the children's HRQoL in terms of physical, psychological and social functioning. Several factors may be accountable for an impairment of the children's HRQoL: firstly, all participating children had been subjected to genital surgery, which is commonly assumed to be emotionally traumatizing (Bracka, 1999). Secondly, children and adolescents with hypospadias are believed to be ashamed of their penile appearance (Mureau et al., 1995b), which could lead to feelings of incompetence, loss of self-esteem and social withdrawal, particularly from the opposite sex. Our results have underlined the importance of a positive penile self-perception for the patients' HRQoL. Thirdly, factors that are potentially affecting the cognitive functions of boys with hypospadias include repeated doctor visits and hospitalizations, which may interrupt regular school attendance. Also, the school environment may become an aversive social setting for some of these boys because the atypical appearance of their penis may contribute to the child becoming the target of teasing by peers. Our results confirmed that the experience of being teased about penile appearance can affect the patients' HRQoL. Ultimately, physical functions of children with hypospadias can be affected by postoperative functional complications such as stenoses and fistulas, which usually cause micturition problems. It is, however, also possible that physical symptoms are psychosomatically triggered by psychological and social factors such as low self-esteem, negative body-image (due to negative penile appraisal) and social withdrawal.

Although mostly parents are interviewed to report on their child's HRQoL in pediatric studies, there are several studies which also focused on self-reported HRQoL of pediatric patients.

Varni, Limbers, and Burwinkle (2007) examined HRQoL in various patient groups (e.g. with diabetes, gastrointestinal conditions, cardiac conditions, asthma, obesity, renal diseases, cancer, rheumatologic conditions, cerebral palsy). Except for children with diabetes, all patient groups showed an impaired self-reported HRQoL in most dimensions (physical, psychological and social functioning). Similar findings have been reported for children with bladder extrophy (Stjernqvist & Kockum, 1999), anorectal malformations (Poley, Stolk, Tibboel, Molenaar, & Busschbach, 2004), spina bifida (Parekh et al., 2006), neurofibromatosis type 1 (Graf, Landolt, Capone Mori, & Boltshauser, 2006), and chronic kidney disease (McKenna et al., 2006). However, there are also studies on pediatric patients that showed a more favorable outcome regarding self-reported HRQoL (e.g. Poretti, Zehnder, Boltshauser, & Grotzer, 2007; Rüth, Landolt, Neuhaus, & Kemper, 2004). Yet, when comparing the results on self-reported HRQoL of the present study with findings of previous studies on pediatric patients with various conditions, it turns out that the impact of hypospadias and its treatment on the patient's well-being must not be underestimated since it seems that hypospadias can affect the patient's HRQoL just like any other serious chronic condition.

In contrast to the patients' ratings, mothers evaluated their children's HRQoL more optimistically and similarly to the mothers of the control group. This divergent rating between children and mothers deserves attention. In a previous study with healthy Dutch children, parents rated their HRQoL also more optimistically than their children (Theunissen et al., 1998). However, mean differences between the ratings were less pronounced and presented only in some domains, similar to the findings in our control group. In contrast to our findings, previous HRQoL-studies among children with chronic diseases reported either a more pessimistically parental view (McKenna et al., 2006; Rüth et al., 2004; Varni et al., 2007) or similar scores between children and parents (Graf et al., 2006). Therefore, one may speculate that mothers of children with hypospadias might psychologically suppress the impact of hypospadias on their child's well-being. This assumption is supported by an observation we have made during the recruitment process. We asked non-participating families on the telephone why they refused to participate in our study. Nine parents of the 25 non-participating families of the hypospadias group answered that they were not willing to deal with their sons' hypospadias (e.g. "We do not want to talk about our son's hypospadias. This has never been an issue"). Another nine families, who had been considering the idea of participating for a considerably length of time, confirmed their participation after several telephone contacts, but they neither kept the

interview appointment nor did they return the questionnaire. We have, however, not made similar observations neither with regard to the control group nor in previous studies on pediatric patients. Moreover, reports of adults show that most parents suppressed their son's hypospadias and kept it secret (Walker, 1998). Unfortunately, due to the lack of previous studies on HRQoL in this group of patients, we are unable to draw comparisons between our results and previous findings. Thus, it is essential to conduct further studies in order to investigate whether the observed discrepancies between child- and mother-reported HRQoL can be confirmed in other samples of children with hypospadias. Should this be the case, it would be important to examine to which extent maternal defense mechanisms actually can be made accountable for the observed discrepancies.

This study also examined predictive values of medical, sociodemographic and psychosocial factors of outcome measures. We hypothesized that more severe hypospadias, higher age at corrective surgery, a higher number of operations, a higher number of days in hospital, a shorter follow-up since the last surgery and a better quality of the surgical result would be associated with a more positive outcome. Furthermore, we postulated that current age is negatively and socioeconomic status as well as Swiss nationality positively associated with the outcome measures. In addition, we expected that patients' who are informed about their hypospadias, who are not ashamed of their hypospadias and who have not been teased about their hypospadias would show a better outcome.

Regarding medical factors, multiple regression analyses revealed that more severe hypospadias predicted both higher mother-reported HRQoL and better psychological adjustment. These findings are contradictory to our hypotheses and do not coincide with results from previous research (Mondaini et al., 2002; Mureau et al., 1997; Purschke & Standke, 1993; Sandberg et al., 1989, 2001). There may be several reasons which could be accountable for these unexpected findings: on the one hand, it is likely that children with more severe hypospadias get more psychosocial support from their families and more medical attendance due to a higher complication rate. On the other hand, it is debatable that patients with more severe hypospadias are less able to deny their penile condition since severe forms of hypospadias are associated with a more atypical appearance of the penis (Duckett, 1998). Hence, they might develop better coping strategies. Landolt, Grubenmann and Meuli (2000) interpreted their results in a similar way. They did not find any differences in psychological adjustment between children with facial burns and children with burns at invisible body locations. They

argued that children with facial burns were forced to deal with their burns since they were not able to hide them. As a consequence, they may have learned to cope with them.
Another medical variable turned out to be an important predictor of gender-role behavior: Younger age at final surgery predicted a more pronounced masculine gender-role behavior. This result confirms our hypothesis and supports the presumption that early surgical treatment may prevent negative effects on the patients' gender development. However, from our point of view, it is debatable whether more feminine behavior actually indicates psychosexual maladjustment given the fact that gender differences in behavior preferences have been decreasing since the issue of gender equality has been raised (Cohen-Kettenis & Pfäfflin, 2003). Also, age at corrective surgery was not associated with HRQoL and psychological adjustment, which we would consider to be more important indicators for the patients'well-being than gender-role behavior.

No other medical factors than those described above turned out to be risk factors for the patients' HRQoL, psychological adjustment and psychosexual development (penile self-perception, gender-role behavior, first sexual experiences, sexual attitude). These findings are in line with previous results (Mondaini et al., 2002; Mureau et al., 1995a, 1995b, 1997) and suggest that objective medical criteria such as the quality of the surgical result are not essential for the patients' well-being. Instead psychosocial factors were found to be important predictors of outcome measures: Consistent with our hypotheses, "being less ashamed of penile appearance" predicted better self-reported HRQoL, and a more positive penile self-perception predicted better mother-reported HRQoL. Furthermore, the patients' experience in having been teased was negatively associated with both mother-reported HRQoL and psychological adjustment. The finding that patients who were informed about their hypospadias did show a more positive penile self-perception also corresponds with our hypotheses and reinforces the claim of support groups and experts in the field of DSD's for age-appropriate information for the patients about their penile condition. Non-disclosure of the child's DSD by medical staff and the child's family is believed to traumatize the patients psychologically and to impede them from developing adequate coping strategies (ISNA, 1994, 2006; Lee, Houk, Ahmed, & Hughes, 2006).

Tenable explanations for the finding that medical variables in comparison to psychosocial factors had little impact on the outcome include the following: on the one hand, our data

might reflect the recent achievements in surgical treatment. Nowadays, it is in fact possible to obtain satisfactory functional and cosmetic results in the vast majority of cases (Duckett, 1998). Numbers of operations and length of hospitalization have been diminishing during the last years (Bracka, 1999). This might have led to medical characteristics becoming less important regarding HRQoL, psychological adjustment and psychosexual development. On the other hand, the positive impact of psychosocial aspects such as age-appropriate information for the patients about their malformation, disclosure of hypospadias in the family, a self-confident attitude towards atypical penile appearance and the ability to assert oneself against teasing peers might outweigh the negative physical consequences of hypospadias. Previous studies on pediatric patients have shown that the children's ability to deal with their chronic condition, their coping behavior as well as family background variables can be more important predictors of the children's HRQoL and psychological adjustment than medical variables (Graf et al., 2006; Landolt et al., 2000; Rüth et al., 2004; Stam, Grootenhuis, Brons, Caron, & Last, 2007; Stam, Grootenhuis, Caron, & Last, 2006; van der Zaag-Loonen, Grootenhuis, Last, & Derkx, 2004).

Regarding sociodemographic risk factors, consistent with our hypothesis, Swiss nationality was a predictor of positive psychological adjustment. Considering that foreign children usually live under more difficult conditions (e.g. less social acceptance) than Swiss children, the result is consistent with our hypothesis. One may also speculate that the association may be caused by cultural differences in coping with hypospadias which may be traced back to cultural differences with respect to general handling of sexuality and genitalia (de Graaf & Rademakers, 2006; Friedrich, Sandfort, Oostveen, & Cohen-Kettenis, 2000; Laumann, Gagnon, Michael, & Michaels, 1994; Laumann et al., 2006). Kuhnle and Krahl (2002) reported that parents in more patriarchal ethnic groups (e. g. Chinese, Muslims) had more difficulties in accepting their child's ambiguous genitalia than parents from less patriarchal societies (e. g. Germans). The authors concluded that the former may feel more threatened by disorders of sex development due to their more rigid traditional gender binary system. Most foreign children in our study originate from countries of Former Yugoslavia, which show a more patriarchal societal structure than Switzerland (Laumann et al., 2006). It might therefore be assumed that parents of children from Former Yugoslavia perceive hypospadias more frequently to be affecting their son's masculinity than Swiss parents and thus have more prob-

lems in dealing with their child's malformation. Such a negative attitude of parents towards hypospadias might have a negative effect on the child's well-being (Sandberg et al., 2001).
A further explanation would be that the association between nationality and psychological adjustment was caused by a lower socioeconomic status of immigrant children. Low income families may experience greater stress in their daily lives than families with higher incomes. However, there were only minor associations between socioeconomic status and the outcome measures and the influence of socioeconomic status was controlled in the multiple regression analyses.

Little is known about ethnic differences in psychological adjustment and HRQoL-outcomes in pediatric patients. Most studies on pediatric patients focused on investigating the influence of socioeconomic status. Meeske, Patel, Palmer, Nelson, and Parow (2007) reported a negative correlation between ethnic minority status and psychosocial functioning and HRQoL in pediatric cancer survivors. In the study of van Dellen et al. (2007), non-immigrant children with asthma showed higher HRQoL-scores than immigrant children with asthma. However, ethnic differences disappeared when socioeconomic status was controlled. With regard to healthy children, results are inconsistent (Achenbach, 1991; Nguyen, Huang, Arganza, & Liao, 2007). Thus, future studies need to clarify the relationships between ethnicity and psychological adjustment and HRQoL, respectively, in both pediatric patients and healthy children.
As mentioned above, the influence of socioeconomic status on children's well-being has been more frequently studied than the impact of nationality. Whereas results of studies on the healthy population indicate that both low parental income and low parental education are risk factors for the children's psychological adjustment and HRQoL (Rüden, Gosch, Rajmil, Bisegger, & Ravens-Sieberer, 2006; Steinhausen, 1987; Varni, Burwinkle, & Seid, 2006), authors of studies on pediatric patients presented inconsistent results (Erickson et al., 2002; Graf et al., 2006; Landolt, Nuoffer, Steinmann, & Superti-Furga, 2002; Landolt, Valsangiacomo, & Latal, in press; Vollrath & Landolt, 2005).

With regard to current age, younger age at assessment showed to be a risk factor for both an impaired child-reported HRQoL and behavioral maladjustment, whereas higher age was negatively associated with penile self-perception. These observations are interesting in that way that boys with hypospadias are assumed to suffer mainly during puberty due to a changing physical self-perception as well as the increasing importance of sexuality (Bracka, 1999). In

concordance with this assumption, our results and previous findings suggest that adolescent patients are less satisfied with penile appearance than younger children (Mureau et al., 1995b). Yet, the finding that current age is positively correlated with both self-reported HRQoL and psychological adjustment is surprising and does not coincide with previous studies (Mondaini et al., 2002; Mureau et al., 1997; Purschke & Standke, 1993; Sandberg et al., 1989, 2001). One plausible explanation for an impaired HRQoL and behavioral maladjustment in younger children might be a shorter follow-up since their last corrective surgery. Assuming that genital surgery is an aversive experience for the patients, it would not be surprising if a longer length of time after surgery had a positive effect on the patients' well-being. In the present study, time of follow-up was positively associated with several dimensions of both self-reported and proxy-reported HRQoL. However, when current age was controlled through partial correlations, the associations between follow-up and the outcome measures lost significance. This suggests that age itself is the more important risk factor for the children's HRQoL and psychological adjustment than time of follow-up. Poley et al. (2004) reported that preschool children with congenital anorectal malformations showed lower HRQoL than school children with congenital anorectal malformations. The authors argued that older children may cope more successfully with their stool difficulties than younger patients. Similarly, older children with hypospadias might deal better with their urinating problems. However, most children of our sample did not have any complications that would have affected micturition. Thus, the role of the patient's age in relation to his psychological well-being needs to be clarified in future studies.

5.2 Methodological Considerations and Limitations

This is the first comprehensive cross-sectional analysis on HRQoL, psychological adjustment, penile self-perception, gender-role behavior, first sexual experiences and sexual attitude of children and adolescents with hypospadias. The strengths of the study include the use of multiple sources of information, the multidimensional and highly standardized assessment of HRQoL and psychological adjustment and the use of multivariate statistics. Furthermore, the study involved a control group equally large to the patient sample and successfully matched regarding current age and age at first operation. Nevertheless, some limitations need to be mentioned: firstly, since the present study bears an exploratory character, we presented a high number of bivariate correlations, which increased the probability of alpha error. However, this problem was countered by conducting multivariate analyses. Secondly, the markedly higher

response rate for the hypospadias patients (study on HRQoL and psychological adjustment: 75.5%; study on psychosexual development: 66.6%) than for the comparison males (study on HRQoL and psychological adjustment: 58.8%; study on psychosexual development: 59.7%) might have biased the results. On the one hand, non-participating hypospadias patients may be particularly ashamed of their condition and the level of HRQoL as well as of psychological and psychosexual adjustment of the participating patients may therefore be overestimated in the present study. On the other hand, the control group is more self-selected for volunteerism due to the lower participation rate. Since research has demonstrated that volunteers are generally better adapted than the general population (Kazdin, 2003), outcome scores of the control subjects might be more likely to be overestimated in comparison to those of hypospadias patients. Thirdly, our sample is small, particularly with regard to the number of adolescents who were interviewed about their first sexual experiences and sexual attitude. Although small sample sizes are typical for studies on pediatric patients with infrequent conditions, this restricts the generizability of our results.

A final methodological consideration refers to the applied measures. Both the TACQOL and the CBCL are generic measures of HRQoL and psychological adjustment, respectively, and may thus lack sensitivity for specific problems of children and adolescents with hypospadias (e. g. micturition problems, sexual anxieties). However, to the best of our knowledge there are no hypospadias-specific measures for HRQoL and psychological adjustment. With regard to psychosexual development, we applied self-developed measures except for gender-role behavior. While the Penile Perception Scale proved to be a reliable measure, the Sexual Attitude Scale showed only a Cronbach's Alpha of .58. Unfortunately, we could not find any other appropriate validated measures of psychosexual development in the German language.

5.3 Conclusions: Directions for future research and clinical implications

The aim of the present study was to shed light on the psychosexual development, HRQoL and psychological adjustment of boys with hypospadias. The study bears an exploratory character and our data point out several issues for future research activities: since it is the first investigation on this subject, our study may be considered to be a basis for further studies conducted in other countries, with larger samples and samples with different age ranges. In particular, prospective studies are necessary to fully understand the factors that may influence long-term outcome and to understand causal relationships between predictors. For instance, the role of the child's current age and age at surgical correction of hypospadias urgently require further

clarification. Furthermore, despite the statistical significance of our findings, multivariate analyses explained only between 26% and 35% of the variability of outcome measures. Thus, variables not included in this study, such as the patient's and parents' coping strategies, may be of particular importance.

In addition, the development and validation of both a hypospadias-specific HRQoL-measure and standardized measures of different constructs of psychosexual development should be taken into consideration. As for HRQoL, such an instrument would ideally assess the boy's perception of his physical (e. g. micturition, sexual function), psychological (e. g. depressive emotions, senses of shame, body image) and social (e. g. social withdrawal, being teased by peers, concealment of hypospadias) functions. Regarding psychosexual development, validated measures of first sexual experiences, sexual behavior habits, sexual attitude and sexual problems have to be developed. Such measures should not only be sensitive to the specific challenges of hypospadias, but should also provide reference data about the psychosexual development of healthy children. To the best of our knowledge, there are excellent evaluated measures of psychosexual differentiation for both healthy children and children with DSD (Meyer-Bahlburg, Sandberg, Yager, Dolezal, & Ehrhardt, 1994). However, validated measures of other constructs of psychosexual development have yet to be developed.

Ultimately, it would be important to compare our results with data from untreated children with hypospadias. Although we did not find any associations between number of operations and the outcome measures, our data do not exclude the possibility of a single penile operation to affect the patients' well-being. Children with hypospadias showed an impaired self-reported HRQoL and multiple regression analysis explained only 29% of the variability in child-reported HRQoL. Therefore, it can be assumed that genital surgery has contributed to the children's impaired HRQoL. The issue of the potential psychological risk of genital surgery should be particularly raised with respect to surgical correction of mild hypospadias, which is exclusively performed due to cosmetic reasons (Duckett, 1998). Support groups have recently announced the need for a reduction in the frequency of genital surgery due to the potential of emotional trauma (ISNA, 2006; M. Walker, personal communication, December 13, 2006). One can therefore speculate that untreated patients would show better psychological adjustment and HRQoL than surgically treated patients. Yet, it is also likely that untreated

patients suffer more often from a negative penile perception due to a more atypical penile appearance, which may have a negative effect on the childrens' psychological well-being.
Unfortunately, there is hardly any data on untreated boys with hypospadias. Mondaini et al. (2002) did not detect any differences in personality profile between six untreated and 36 surgically treated adolescents with hypospadias but reported that the untreated boys were sexually more inhibited than the treated boys. Purschke und Standke (1993) presented data on eight untreated children with hypospadias. The latter showed higher rates of psychological maladjustment than the 39 boys after surgical treatment. However, the examined groups of untreated patients in these two studies were much too small to generalize results. In 2003, the US self-help group Hypospadias and Epispadias Association conducted an online survey about the sexual lives of men with hypospadias. Overall, men who did not undergo any surgery had fewer sexual problems and a more positive penile self-perception than men who were surgically treated (Hypospadias and Epispadias Association, 2003). However since severity of hypospadias was not statistically controlled and most untreated participants had mild hypospadias, the observed group differences might rather be a consequence of severity of hypospadias than of genital surgery.

Our findings also implicate some consequences for clinical management. In order to guarantee a comprehensive treatment for children with hypospadias, not only surgical results should be carefully evaluated, but also the childrens' HRQoL. In doing so, both children and parents should be asked, since the results showed that proxy- and self-perception of HRQoL can considerably diverge. Surgeons need to be aware that HRQoL and psychological adjustment of children with hypospadias might be impaired by a negative penile self-perception, sense of shame or teasing remarks from peers. As previous studies have shown that social skill training can improve social functioning and quality of life in pediatric patients (Barakat et al., 2003; Grey, Boland, Davidson, & Tamborlane, 2000; Varni, Katz, Colegrove, & Dolgin, 1993), it can be expected that psychological support may also help hypospadias patients to better accept their penile appearance and to defend themselves against teasing remarks from their peers.
With regard to psychosexual development, on the one hand, our results suggest that boys with hypospadias may show a psychosexual development similar to healthy boys. On the other hand, they also indicate that puberty might be a critical time for patients. Therefore they may benefit from regular urological follow-ups until young adulthood. Apart from urological ex-

aminations, the patients should be asked how they feel about sexual matters and also, they should be provided with age-appropriate information about their penile condition. Moreover, surgeons must be aware that the objective surgical outcome and severity of hypospadias might be less important for the patients' psychosexual development during childhood and adolescence than psychosocial aspects. Our results, however, indicate that the later corrective surgery is completed the more likely it is that patients become more insecure regarding gender-role behavior.

In summary, the most central findings of this Ph.D.-project are that self-reported HRQoL of children and adolescents with hypospadias can be impaired, although psychological adjustment and psychosexual development seem to be similar to healthy children. In addition, this study underlined the importance of psychosocial aspects for the patients' well-being: overall, psychosocial factors (e.g. penile self-perception) have shown to be more important predictors of outcome than medical aspects (e. g. quality of surgical outcome). This is essential since to date the treatment concept for hypospadias patients has been focusing on optimizing surgical results rather than on psychological support of the patients.

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6 Appendix

6.1 Appendix A: Invitation for Study Participation

KINDERSPITAL ZÜRICH
Universitäts-Kinderklinik · Eleonoren-Stiftung

CHIRURGISCHE KLINIK UND POLIKLINIK
Studie zum Langzeitverlauf bei Kindern und Jugendlichen nach Hypospadiekorrektur

Name
Adresse

Zürich, Datum

Einladung zur Teilnahme an der Studie
Langzeitverlauf bei Kindern und Jugendlichen nach Hypospadiekorrektur

Lieber *Vorname des Kindes*

Sehr geehrte Familie *Name*

Vor 15 Jahren wurde *Vorname des Kindes* wegen einer Hypospadie am Kinderspital Zürich operiert. Wir hoffen, dass *Vorname des Kindes* bezüglich der Operation beschwerdefrei ist.

Obwohl Hypospadien relativ häufig auftreten und auch operiert werden, ist bis heute weitgehend unbekannt, welche Auswirkungen eine Hypospadie auf die Lebensqualität und die Entwicklung von Jugendlichen hat. Neuere Operationstechniken erlauben es, Hypospadien zusehends früher zu operieren. Jedoch ist bisher ungeklärt, in welchem Alter der ideale Zeitpunkt für eine Operation aus psychologischer Sicht liegt.

Um all diese Fragen zu beantworten, wird in diesem Jahr eine Studie durchgeführt. Dazu möchten wir ehemalige Patienten und deren Eltern zu einer Befragung einladen. Daher ersuchen wir auch *Vorname des Kindes* und Sie als Eltern höflichst, an unserer Untersuchung teilzunehmen. Mit Ihrer Erfahrung könnten Sie uns helfen, die Behandlung für zukünftige Patienten zu verbessern.

Gerne möchten wir Sie in Form eines schriftlichen Fragebogens und im Rahmen eines Interviews zur Entwicklung befragen. Anschliessend möchten wir *Vorname des Kindes* untersuchen, so wie wir das schon in der letzen Poliklinik-Sprechstunde gemacht haben. Die Untersuchungen werden nicht schmerzhaft sein, und es erfolgt auch keine Blutentnahme. Wir wären sehr froh, wenn Sie für die Untersuchung in die chirurgische Poliklinik kommen könnten.

Transportkosten können wir Ihnen zum Preis der öffentlichen Verkehrsmittel zurückerstatten. Sollte der Weg ans Kinderspital für Sie schwierig sein, so können wir auf Ihren Wunsch auch zu Ihnen nach Hause kommen. Da es sich um eine Studie handelt, entstehen für Sie oder für Ihre Krankenkasse keine Kosten.

Oft haben Eltern und Jungen Jahre nach einer Hypospadieoperation nie gestellte oder neue Fragen, zum Beispiel auch zur Sexualität. Selbstverständlich haben wir im Rahmen der Untersuchung auch die Möglichkeit, auf Ihre entsprechenden Fragen einzugehen.

Wir wären Ihnen sehr dankbar, wenn Sie sich zu einer Teilnahme an der Studie entschliessen würden. Sie helfen damit, die chirurgische Behandlung von Kindern mit Hypospadien in der Zukunft weiter zu verbessern. Selbstverständlich ist die Teilnahme an der Studie freiwillig und eine Nicht-Teilnahme hat keinerlei Auswirkungen auf zukünftige Behandlungen bei Ihrem Kind. Auch wenn Sie nicht an der Studie teilnehmen, sind wir gerne bereit, Ihre Fragen zur Hypospadie von *Vorname des Kindes* im Rahmen einer normalen Konsultation zu beantworten.

Falls Sie an der Studie teilnehmen möchten, bitten wir Sie, das beigelegte Einverständnisformular zu unterzeichnen und im frankierten Umschlag bis zum 20. 7. 06 an uns zu retournieren. Wir werden Sie darauf telefonisch kontaktieren, um mit Ihnen einen Termin für die Untersuchung zu vereinbaren. Falls der Termin in die Schul- oder Arbeitszeit Ihres Sohnes fallen sollte, schreiben wir gerne eine Bestätigung für den/die Lehrer/in oder den/die Arbeitgeber/in.

Bei Fragen steht Ihnen unsere wissenschaftliche Mitarbeiterin, Frau lic. phil. Verena Schönbucher telefonisch, schriftlich oder per E-Mail zur Verfügung (Tel.: 044 266 80 23; E-Mail: Verena.Schoenbucher@kispi.unizh.ch).

Mit freundlichen Grüssen

Dr. Daniel M. Weber Studienleiter Oberarzt Chirurgische Klinik	PD Dr. Rita Gobet Leitende Ärztin Urologie	lic. phil. Verena Schönbucher Wissenschaftliche Mitarbeiterin

6.2 Appendix B: Informed Consent

KINDERSPITAL ZÜRICH
Universitäts-Kinderklinik · Eleonoren-Stiftung

CHIRURGISCHE KLINIK UND POLIKLINIK
Studie zum Langzeitverlauf bei Kindern und Jugendlichen nach Hypospadiekorrektur

Einverständniserklärung der Eltern

Name des Kindes:

Name der Eltern:

Adresse:

Telefonnummer:

Wir erklären uns einverstanden mit der Teilnahme an der wissenschaftlichen Studie zum Langzeitverlauf bei Kindern und Jugendlichen nach einer Hypospadiekorrektur. Wir haben den entsprechenden Informationsbrief gelesen und wissen, dass unser Kind von einer Fachperson persönlich zu seiner Lebensqualität, seinem Befinden, seinen Operationen und zu seiner psychosexuellen Entwicklung befragt wird und anschliessend medizinisch untersucht wird. Wir haben unser Kind über die Studie informiert. Als Eltern (Mutter und/oder Vater) erklären wir uns bereit, einen Fragebogen zur Situation unseres Kindes und unserer Familie auszufüllen.
Wir nehmen Kenntnis, dass alle am Projekt beteiligten Personen der Schweigepflicht unterstehen und die erhobenen Daten nur anonymisiert verwendet und bearbeitet werden.
Wir haben jederzeit das Recht, die Teilnahme an der Studie abzubrechen, wenn wir dies möchten. Die Teilnahme an der Studie ist freiwillig. Eine Nicht-Teilnahme hat keinerlei Auswirkungen auf die Behandlung unseres Kindes.

Datum:

Unterschrift der Eltern:

6.3 Appendix C: Assessment Form for Medical Data

ID-Nummer:……

Datenerfassungsblatt Medizinische Daten

Name:

Vorname:

Geburtsdatum:

Nationalität:

Sprache:

Konfession:

Vater (Name, JG, Beruf):

Mutter (Name, JG, Beruf):

Familienanamnese: □ bland □ k.A. □ pos

Nebendiagnosen:

Diagnose intraoperativ:
Hypospadie: □ glanulär □ coronar □subcor. □ penil □ penoscrot

Chorda □ ja □ nein □ k. A.

Erstoperation
Hosp. von/bis:

Was: □ Mathieu □ MAGPI □ andere: ……

Wo: □ Kispi □ Ausland □ CH andere: ……

Operateur: □ Chef □ LA □ OA □ AA

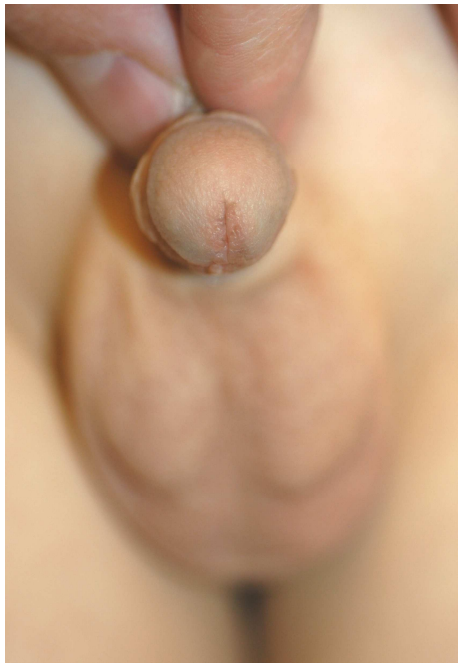
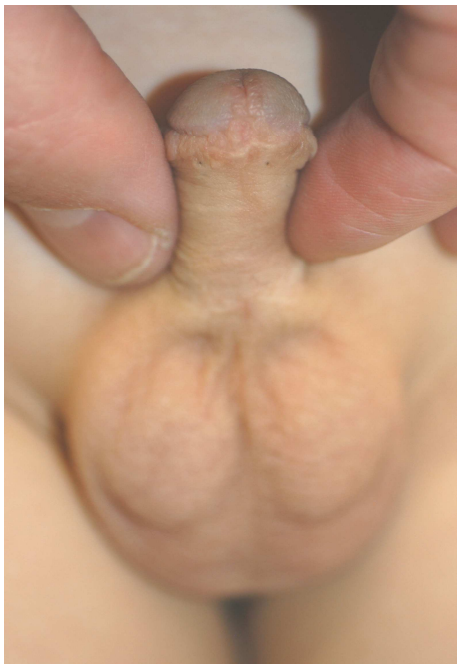
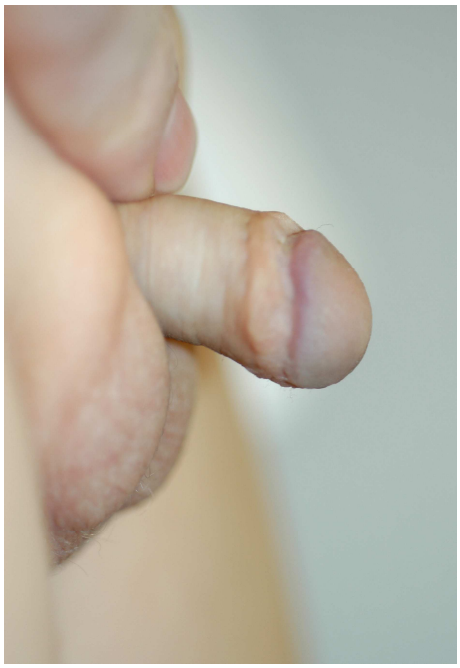
Katheter: □ transurethral □ suprapubisch Anzahl Tage:……..

Wieso dann/so spät:

Komplikationen (was, wann):

6.4 Appendix D: Assessment Form for Quality of Surgical Results

Patientennr.: Rater:

PPPS	Very satisfied	satisfied	dissatisfied	Very dissatisfied
Meatal position and shape	☐	☐	☐	☐
Glans shape	☐	☐	☐	☐
Shape of penile skin	☐	☐	☐	☐
Penile axis	☐	☐	☐	☐
General cosmetic appearance	☐	☐	☐	☐

6.5 Appendix E: Children's Interview

Interview für das Kind

ID-Nummer:…....

Name und Vorname des Kindes:……..

Datum des Interviews:……..

Durchführungsort des Interviews: □ Kinderspital Zürich

□ zu Hause

Vor der Durchführung des Interviews soll mit dem Kind einige Minuten Kontakt aufgenommen werden. Die Interviewerin/der Interviewer stellt sich zuerst selber vor und erklärt, wieso sie/er hier ist. Das Kind soll wenn möglich alleine, d.h. ohne Beisein der Mutter befragt werden. Falls die Mutter auf Wunsch des Kindes im selben Raum bleibt, sollte sie etwas abseits sitzen und mit etwas beschäftigt sein (Fragebogen ausfüllen).

Ich möchte nun mit Dir über verschiedene Themen sprechen: z. B. über deine Freundschaften, darüber wie es Dir geht und über die Operation/en, die an Deinem Penis/Schnäbi gemacht worden ist/sind. (ab 12 J.: und über Sexualität). Dazu stelle ich Dir Fragen. Bei den meisten Fragen werde ich Dir verschiedene Möglichkeiten für Deine Antwort vorlesen. Du sollst mir jeweils sagen, welche Antwort am ehesten auf Dich zutrifft. Dabei gibt es keine richtigen und falschen Antworten, Du sollst einfach nur das sagen, was zu Dir passt. Bitte sage mir, wenn Du eine Frage oder eine Antwort nicht verstehst, dann werde ich dir diese nochmals vorlesen oder besser erklären.

Beginnen wir mit der Schule…

1. In welche Klasse gehst Du?
Ich gehe in die _____ Klasse (Weiss nicht □(9))

Fragen zu Freundschaften

Als nächstes kommen ein paar Fragen zu Deinen Freundschaften.

2. Wieviele Freunde und Freundinnen hast Du?

□(3) Ich habe mehr Freunde und Freundinnen als andere Kinder/Jugendliche

□(2) Ich habe ähnlich viele Freunde und Freundinnen wie andere Kinder/Jugendliche

□(1) Ich habe weniger Freunde und Freundinnen als andere Kinder/Jugendliche

□(0) Ich habe keine Freunde und Freundinnen ***→ bitte weiter mit Frage 5*** (Weiss nicht □(9))

3. Mit wem schliesst Du am häufigsten Freundschaften?

□(0) mit Mädchen □(2) mit Jungen □(1) mit Mädchen und Jungen (Weiss nicht □(9))

4. Hast Du einen besten Freund oder eine beste Freundin?

□(1) ja □(0) nein (Weiss nicht □(9))

5. Wie oft kommt es vor, dass Du von anderen Kindern gehänselt (geärgert) wirst?

□(0) nie □(1) manchmal □(2) oft (Weiss nicht □(9))

Gibt es noch etwas Wichtiges, das Du zu Deinen Freunden und Freundinnen loswerden möchtest?

_____

_____

_____

_____

_____

Fragen zur Lebensqualität

Nun möchte ich gerne wissen, wie es Dir in den letzten 2 Wochen <öpe zwei Wuche> ging. Bei den folgenden Fragen hast Du wieder verschiedene Antwortmöglichkeiten. Die Antwortmöglichkeiten hab ich diesmal auf diese Streifen aufgeschrieben. Sie lauten…(Antworten vorlesen) Wähle immer diejenige Antwort aus, die am besten zu Dir passt.

6. Hattest Du gelegentlich Ohren- oder Halsschmerzen?

- nie ☐ (4)
- manchmal ☐
- oft ☐

} Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

7. Hattest Du gelegentlich Magen- oder Bauchschmerzen?

- nie ☐ (4)
- manchmal ☐
- oft ☐

} Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

8. Hattest Du gelegentlich Kopfschmerzen?

- nie ☐ (4)
- manchmal ☐
- oft ☐

} Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

9. War es Dir gelegentlich schwindlig?

- nie ☐ (4)
- manchmal ☐
- oft ☐

} Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

10. War es Dir gelegentlich übel?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

11. Warst Du müde?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

12. Warst Du träge?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

13. Warst Du benommen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

14. Hattest Du andere Schmerzen oder Beschwerden?

Welche?_____

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Hattest Du gelegentlich…

15. Mühe mit dem Gehen?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

16. Mühe mit dem Rennen?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

17. Mühe mit dem Stehen?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

18. Mühe mit dem Heruntergehen von Treppen?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

19. Mühe mit dem Spielen?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

20. Mühe mit dem langen Gehen oder Laufen, mit der Ausdauer?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

21. Mühe mit dem Halten Deines Gleichgewichts?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

22. Mühe, Dinge geschickt und schnell zu erledigen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Hattest Du gelegentlich…

23. Mühe selbständig zur Schule zu gehen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

24. Mühe, Dich selbst zu waschen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

25. Mühe, Dich selbst anzukleiden?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

26. Mühe, selbst zum WC zu gehen

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

27. Mühe, selbst zu essen und zu trinken?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

28. Mühe selbst Sport zu treiben oder draussen zu spielen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

29. Mühe selbst Hobbys zu treiben?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

30. Mühe mit dem Fahrradfahren?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Hattest Du gelegentlich…

31. Mühe mit dem Aufpassen oder Konzentrieren?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

32. Mühe mit dem Begreifen der Schularbeit?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

33. Mühe mit dem Begreifen von dem was andere gesagt haben?

nie ☐ (4)

manchmal ☐

oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

34. Mühe mit dem Rechnen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

35. Mühe mit dem Lesen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

36. Mühe mit dem Schreiben?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

37. Mühe mit dem Lernen?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

38. Mühe, die richtigen Worte zu finden?

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Mit anderen Kindern und Deinen Eltern in den vergangenen zwei Wochen:
Denke jeweils kurz darüber nach, wie es in den letzten zwei Wochen war..

39. Ich konnte mit anderen Kindern gut spielen oder sprechen.

oft ☐ (4)
manchmal ☐
nie ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

40. Ich konnte bei anderen Kindern mich selbst behaupten.

oft ☐ (4)
manchmal ☐
nie ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

41. Andere Kinder baten mich mitzuspielen.

oft ☐ (4)
manchmal ☐
nie ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

42. Ich fühlte mich bei anderen Kindern wohl.

oft ☐ (4)
manchmal ☐
nie ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

43. Ich konnte mit meinen Eltern gut spielen oder sprechen.

oft ☐ (4)
manchmal ☐
nie ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

44. Meinen Eltern gegenüber war ich schweigsam und still.

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

45. Ich war meinen Eltern gegenüber unruhig oder ungeduldig.

nie ☐ (4)
manchmal ☐
oft ☐

Ich fühlte mich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

46.	Ich war meinen Eltern gegenüber aufsässig.	nie	☐ (4)			
		manchmal	☐	} Ich fühlte mich dabei:		
		oft	☐			
				☐ (3) (sehr) gut	☐ (2) nicht so gut	☐ (1) ziemlich schlecht	☐ (0) schlecht

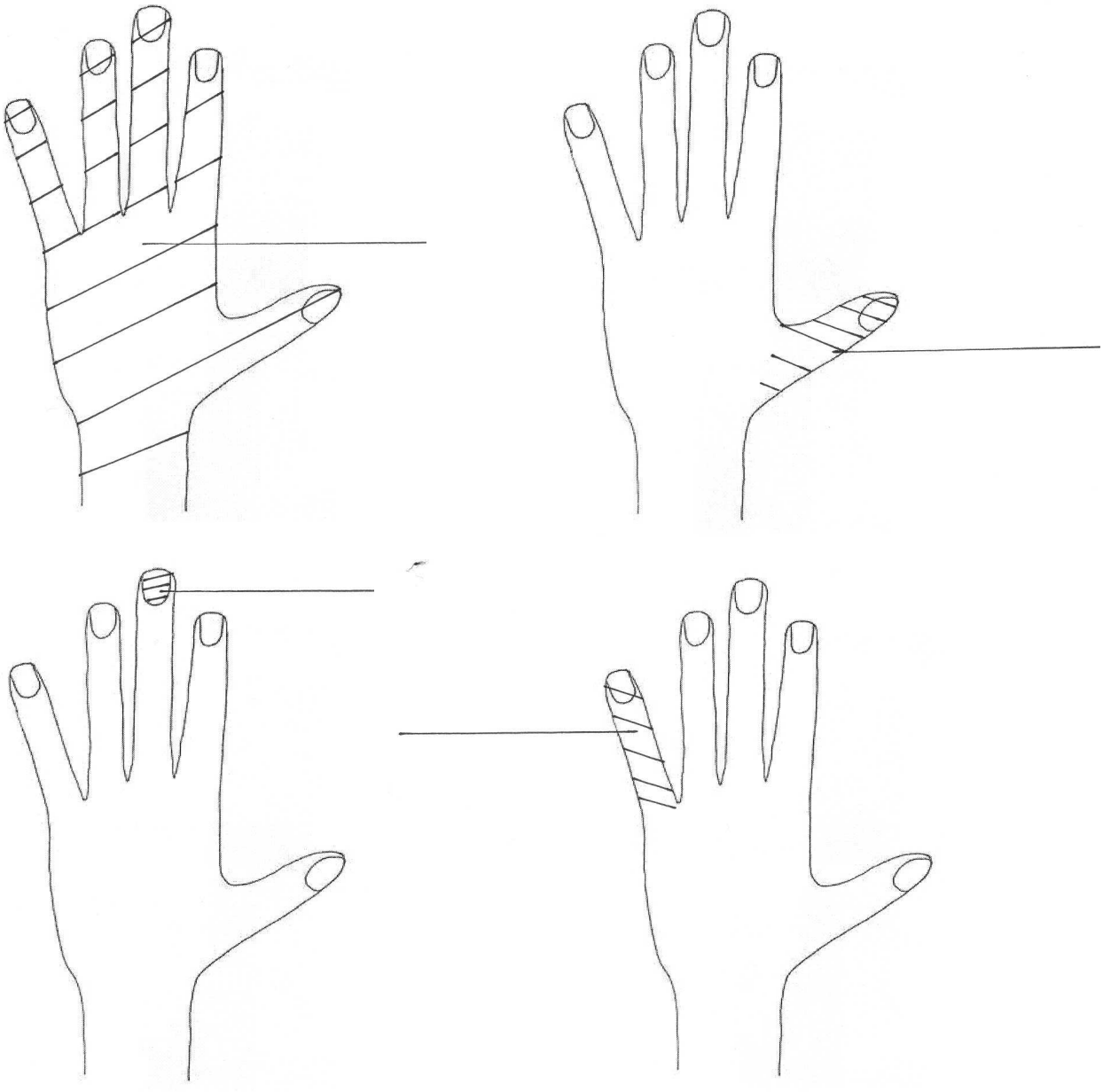
Ich fühlte mich in den letzten zwei Wochen....

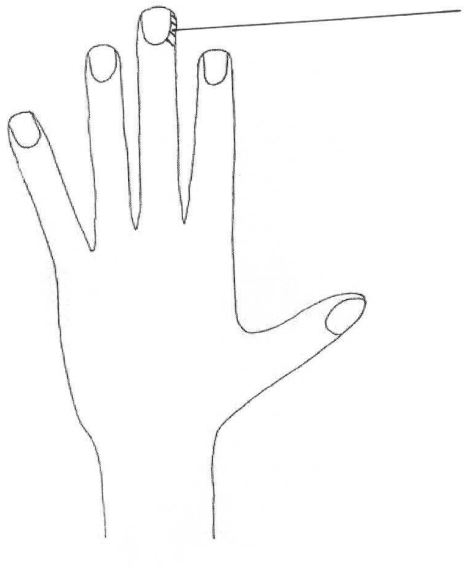
		nie	manchmal	oft
47.	fröhlich……..	☐(0)	☐(1)	☐(2)
48.	betrübt…….	☐(2)	☐(1)	☐(0)
49.	vergnügt…….	☐(0)	☐ (1)	☐(2)
50.	böse…….	☐(2)	☐(1)	☐(0)
51.	zufrieden…….	☐(0)	☐(1)	☐(2)
52.	besorgt……	☐(2)	☐(1)	☐(0)
53.	überschwänglich…….	☐(0)	☐(1)	☐(2)
54.	trübsinnig……	☐(2)	☐(1)	☐(0)
55.	entspannt…….	☐(0)	☐(1)	☐(2)
56.	aggressiv…….	☐(2)	☐(1)	☐(0)
57.	glücklich…….	☐(0)	☐(1)	☐(2)
58.	wütend……	☐(2)	☐(1)	☐(0)
59.	selbstsicher…….	☐(0)	☐(1)	☐(2)
60.	eifersüchtig…….	☐(2)	☐(1)	☐(0)
61.	froh…….	☐(0)	☐(1)	☐(2)
62.	ängstlich…….	☐(2)	☐(1)	☐(0)

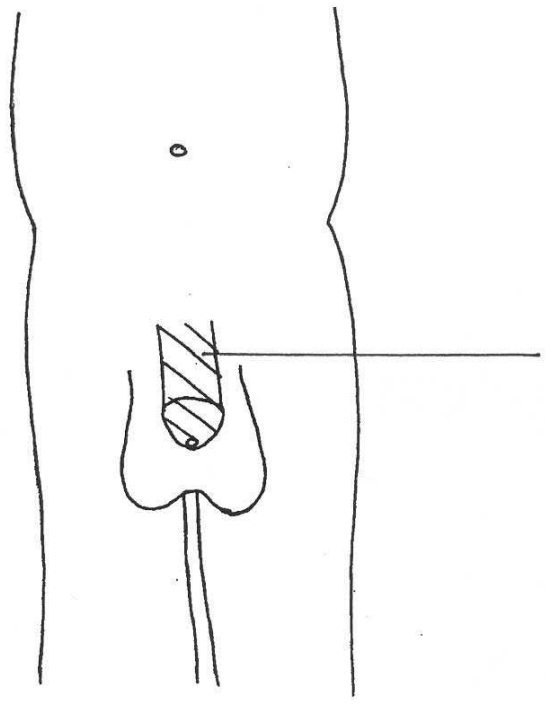
Fragen zum Krankheitskonzept

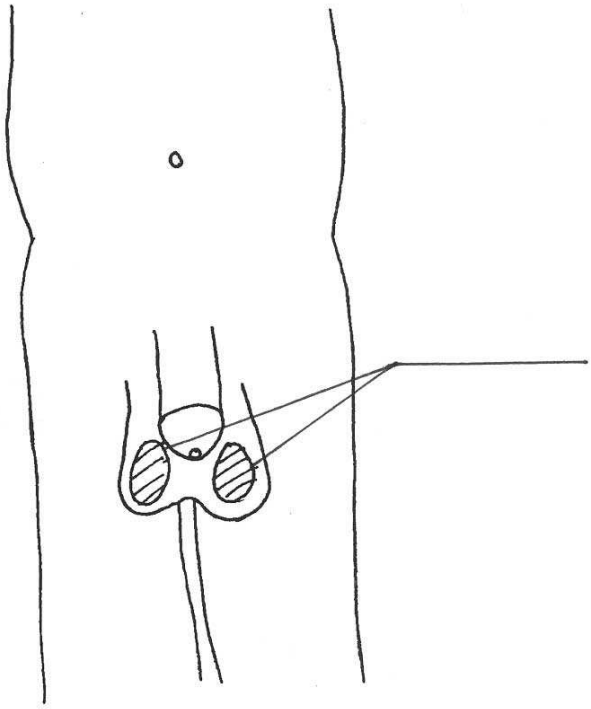
So jetzt hast Du mir einiges darüber erzählt, wie es Dir geht. Nun kommen wir langsam zu Deiner/n Operation/en (nur, wenn aufgeklärt). Davor möchte ich mit Dir aber noch kurz über die männlichen Geschlechtsteile sprechen (jüngere: über Körperteile sprechen, die nur Jungen und Männer haben, z. B. der Penis/das Schnäbi). Zuerst gebe ich Dir ein Blatt mit Zeichnungen von Körperteilen. Ich möchte, dass Du mir jeweils den Namen der schraffierten Körperteile nennst. (Je nach Alter Aufgabe genauer erklären). Wenn Du nicht alle Körperteile benennen kannst, werde ich Dir dabei helfen. Versuche es aber zuerst alleine.

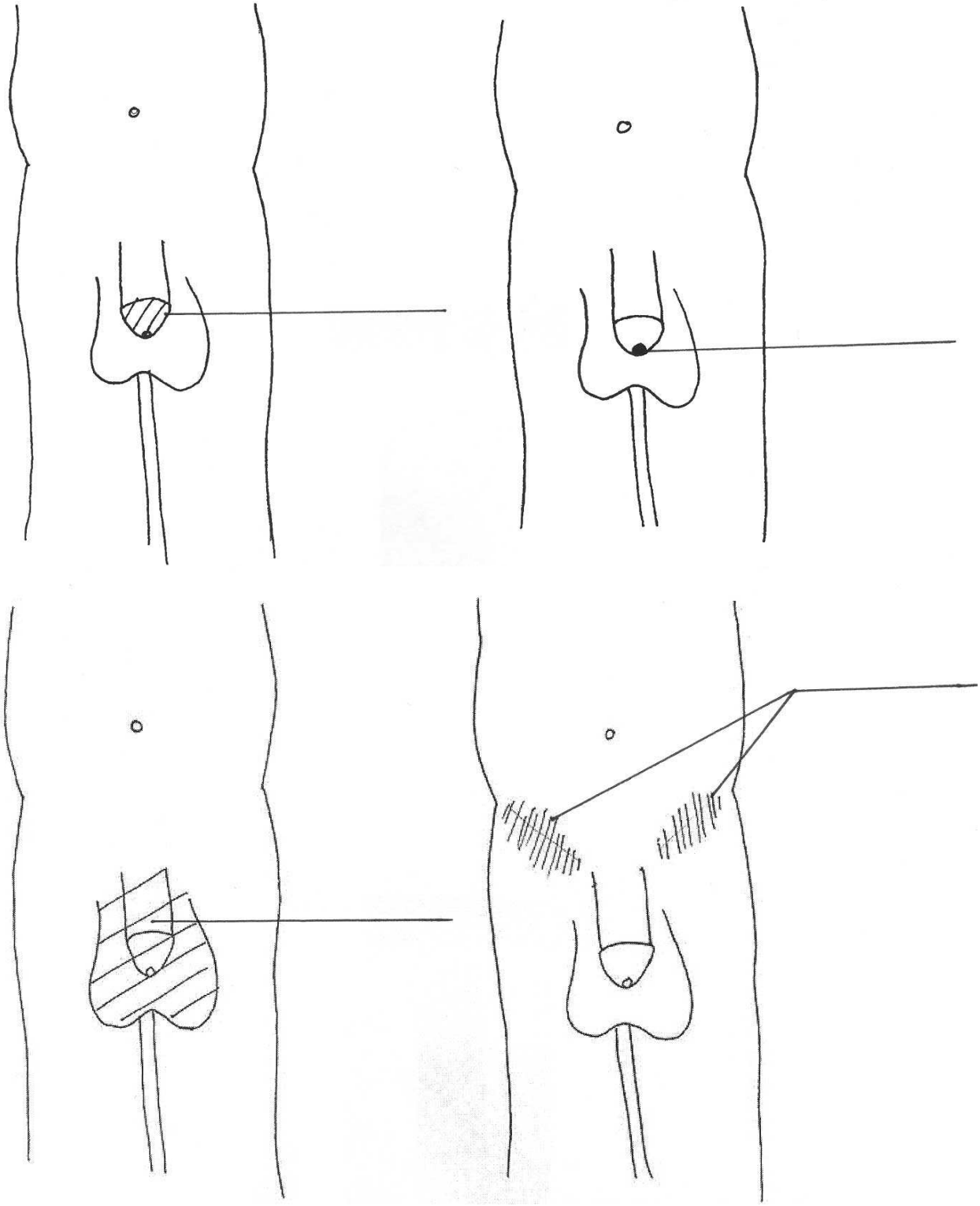
63.











Nun, das hast Du gut gemacht. Jetzt möchte ich etwas über Dein Schnäbi/Deinen Penis und Deine Operation/en sprechen (im Folgenden immer Name auf Skizze verwenden).

64. Du bist an Deinem Penis/Schnäbi operiert worden. Kannst Du mir sagen, warum?

65. Hast Du einen Namen dafür (warum Du an Deinem Penis/Schnäbi operiert worden bist)?

Fragen zur Diagnose und der/n Operation/en

66. Wie lange weißt Du schon, dass Du an Deinem Penis operiert worden bist (Anzahl Jahre)?

_____ (Weiss nicht □(9))

67. Hast Du die Informationen, die Du zu Deiner/n Hypospadie/Operation/en erhalten hast, verstanden?

□(3) Ich habe das Gefühl, die Informationen vollständig verstanden zu haben.

□(2) Ich habe das Gefühl, einige der Informationen verstanden zu haben.

□(1) Ich habe das Gefühl, die Informationen nicht verstanden zu haben.

□(0) Ich kann mich nicht mehr daran erinnern. (Weiss nicht □(9))

68. Hättest Du gerne mehr Informationen zu Deiner Hypospadie/Deiner/n Operation/en?

□(1) ja □(0) nein (Weiss nicht □(9))

Wenn Du Fragen zu Deiner Hypospadie hast, kannst Du diese in der anschliessenden Untersuchung dem Arzt/der Ärztin stellen.

69. Erinnerst Du Dich an Deinen Spitalaufenthalt als Du (das letzte Mal) an deinem Schnäbi/Penis (wegen Deiner Hypospadie) operiert worden bist?

□(1) ja □(0) nein (Weiss nicht □(9))

→ ***Dann werde ich Dich nicht weiter zu Deinem Spitalaufenthalt befragen. Ich stelle Dir aber noch ein paar Fragen zu Deinem operierten Schnäbi/Penis → weiter mit Frage 80***

70. Hat Dein Penis/Schnäbi nach der Operation so ausgesehen wie Du es Dir vorgestellt hast?

□(1) ja □(0) nein (Weiss nicht □(9))

→ weiter mit Frage 72

71. Was sah an Deinem Penis/Schnäbi nach der Operation anders aus als Du Dir vorgestellt hast?

72. Findest Du Die Operation/en hat/haben Verbesserungen an Deinem Penis/Schnäbi gebracht?

☐ ja, **welche Verbesserungen?** *(Mehrfachnennungen möglich)* ***Ich nenne ein paar Dinge, die sich verbessert haben könnten und Du sagst mir, ob sich diese Dinge wirklich verbessert haben.***

☐(1) die Länge meines Penis/Schnäbi
☐(2) die Position und Form der Harnröhrenöffnung
☐(3) die Form der Eichel
☐(4) die Form der Penishaut
☐ (5) die Achse des Penis (ob der errigierte Penis gerade ist)
☐(6) (Fehlen der) Vorhaut (=Haut um die Eichel des Penis)
☐(7) Gesamteindruck des Penis (Aussehen des Penis insgesamt)
☐(8) etwas anderes, was?_____

☐(0) nein (Weiss nicht □(9))

73. Findest Du Die Operation/en hat/haben Verschlechterungen an Deinem Penis/Schnäbi gebracht?

☐ ja, **welche Verschlechterungen?** *(Mehrfachnennungen möglich)* ***Ich nenne ein paar Dinge, die sich verschlechtert haben könnten und Du sagst mir, ob sich diese Dinge wirklich verschlechtert haben.***

☐(1) die Länge meines Penis/Schnäbi
☐(2) die Position und Form der Harnröhrenöffnung
☐(3) die Form der Eichel
☐(4) die Form der Penishaut
☐(5) die Achse des Penis (ob der errigierte Penis gerade ist)
☐(6) (Fehlen der) Vorhaut (=Haut um die Eichel des Penis)
☐(7) Gesamteindruck des Penis (Aussehen des Penis insgesamt)
☐(8) etwas anderes, was?_____

☐(0) nein (Weiss nicht □(9))

74. Wie gut haben Dich Deine Eltern darüber informiert, wie Dein Penis/Schnäbi nach der Operation aussehen würde?

☐(3) (sehr) gut ☐(2) nicht so gut ☐(1) ziemlich schlecht ☐(0) schlecht

(□9 weiss nicht)

75. Wie gut haben Dich die Ärzte darüber informiert, wie Dein Penis/Schnäbi nach der Operation aussehen würde?

☐(3) (sehr) gut ☐(2) nicht so gut ☐(1) ziemlich schlecht ☐(0) schlecht

(□9 weiss nicht)

76. Wie gut haben Dich Deine Eltern auf die Operation vorbereitet? (Dir gesagt, was gemacht wird, wie lange Du im Spital bleiben musst etc.)

☐(3) (sehr) gut ☐(2) nicht so gut ☐(1) ziemlich schlecht ☐(0) schlecht

(□9 weiss nicht)

77. Wie gut haben Dich die Ärzte auf die Operation vorbereitet? (Dir gesagt, was gemacht wird, wie lange Du im Spital bleiben musst etc.)

☐(3) (sehr) gut ☐(2) nicht so gut ☐(1) ziemlich schlecht ☐(0) schlecht

(□9 weiss nicht)

78. Viele Kinder haben vor einer Operation Angst.
a) Wie stark war Deine Angst vor der Operation an Deinem Penis?

☐(4) sehr stark ☐(3) stark ☐(2) mittel ☐(1) ein wenig ☐(0) überhaupt nicht

(□9 weiss nicht)

b) Wovor hattest Du Angst?_____

(□9 weiss nicht)

79. Wie stark waren Deine Schmerzen nach der Operation?

☐(4) sehr stark ☐(3) stark ☐(2) mittel ☐(1) ein wenig ☐(0) überhaupt nicht

(□9 weiss nicht)

80. Wie findest Du die Ergebnisse Deiner Operation aus heutiger Sicht?

☐(3) (sehr) gut ☐(2) nicht so gut ☐(1) ziemlich schlecht ☐(0) schlecht

(□9 weiss nicht)

81. Wie stark stören Dich Deine Narben am Penis?

☐(4) sehr stark ☐(3) stark ☐(2) mittel ☐(1) ein wenig ☐(0) überhaupt nicht

(□9 weiss nicht)

82. Wie stark stört es Dich, dass Dein Penis beschnitten ist (Vorhaut fehlt)?

☐(4) sehr stark ☐(3) stark ☐(2) mittel ☐(1) ein wenig ☐(0) überhaupt nicht

(□9 weiss nicht)

Möchtest Du noch etwas Wichtiges zu Deiner Hypospadie oder Deiner/n Operation/en sagen? Wenn Du Fragen zu der/n Operation/en hast, kannst Du diese in der anschliessenden Untersuchung dem Arzt/der Ärztin stellen.

_____

_____

_____

Fragen zum Umgang mit der Hypospadie und der Wahrnehmung des Penis

Nun möchte ich Dir ein paar Fragen dazu stellen, wie Du mit Deiner Hypospadie umgehst, z. B. wie es Dir beim Ausziehen vor anderen geht. Ebenfalls möchte ich wissen, was Du über das Aussehen Deines Penis/Schnäbi denkst.

83. ***Ich lese Dir nun ein paar Aussagen vor. Bitte sage mir, wie sehr die Aussagen auf Dich zutreffen/zu Dir passen. Die Antwortmöglichkeiten hab ich auf diese Streifen aufgeschrieben. Sie lauten…(Antworten vorlesen). Wähle immer diejenige Antwort aus, die am besten zu Dir passt. (Codierung: ☺: trifft zu 😐: trifft etwas zu ☹: trifft nicht zu) (Beispiel: Ich schaue gerne fern. Wie gut passt der Satz zu dir? Passt, passt ein bisschen/manchmal zu mir, passt nicht zu mir)***

		trifft zu	**trifft etwas zu**	**trifft nicht zu**
a	Meine Eltern sprechen mit mir über meine Hypospadie/meine Operation/en.	☐ (0)	☐ (1)	☐ (2)
b	Ich schäme mich wegen meines Penis.	☐ (2)	☐ (1)	☐ (0)
c	Ich rede mit meinen Freunden und Freundinnen über meine Hypospadie/meine Operation/en.	☐ (0)	☐ (1)	☐ (2)
d	Ich meide öffentliche Duschen/Garderoben (z. B. in der Schule), so dass niemand sieht, dass ich eine Hypospadie habe/dass ich am Penis operiert worden bin.	☐ (2)	☐ (1)	☐ (0)
e	Mir ist es egal, wenn andere Personen in öffentlichen Duschen/Garderoben meine Hypospadie/meinen Penis wahrnehmen.	☐ (0)	☐ (1)	☐ (2)
f	Es kam schon vor, dass jemand mich wegen des Aussehens meines Penis/Schnäbis ausgelacht hat.	☐ (2)	☐ (1)	☐ (0)
g	Ich bin mir bewusst, dass mein Penis anders aussieht als derjenige von anderen Jungs.	☐ (2)	☐ (1)	☐ (0)

84. ***Ich nenne dir im Folgenden verschiedene Eigenschaften deines Penis. Bitte sage mir, wie zufrieden Du mit den entsprechenden Eigenschaften bist. Es gibt immer 4 Antwortmöglichkeiten: sehr zufrieden, zufrieden, unzufrieden, sehr unzufrieden. Ich habe sie Dir wieder auf einen Streifen geschrieben.***

		sehr zufrieden	zufrieden	un-zufrieden	sehr unzufrieden
a	Penislänge	☐(3)	☐(2)	☐ (1)	☐(0)
b	Position und Form der Harnröhrenöffnung	☐(3)	☐(2)	☐ (1)	☐(0)
c	Form der Eichel	☐(3)	☐(2)	☐ (1)	☐(0)
d	Form der Penishaut	☐(3)	☐(2)	☐ (1)	☐(0)
e	Achse des Penis (ob der errigierte Penis gerade ist)	☐(3)	☐(2)	☐ (1)	☐(0)
f	Fehlen der Vorhaut	☐(3)	☐(2)	☐ (1)	☐(0)
g	Gesamteindruck des Penis	☐(3)	☐(2)	☐ (1)	☐(0)

85. Angenommen, eine Operation könnte das Aussehen Deines Penis/Schnäbis verbessern, würdest Du einer solchen Operation zustimmen?

☐(2) ja ☐(0) nein ☐(1) bin mir unsicher (Weiss nicht □(9))

Möchtest Du sonst noch etwas zu Deinem Penis oder Deiner Hypospadie sagen?

Fragen zum Körper

Nun haben wir über das Aussehen Deines Penis gesprochen. Nun möchte ich auch noch wissen, wie Du zu Deinem Körper im Allgemeinen stehst.
Im Folgenden werde ich Aussagen zu Deinem Körper machen. Bitte sage mir, wie sehr die Aussagen auf Dich zutreffen/zu Dir passen. Die Antwortmöglichkeiten habe ich auf diese Streifen aufgeschrieben. Sie lauten…(Antworten vorlesen). Wähle immer diejenige Antwort aus, die am besten zu Dir passt. (Codierung: ☺: trifft zu 😐: trifft etwas zu ☹: trifft nicht zu) (Beispiel: Ich schaue gerne fern. Wie gut passt der Satz zu dir? Passt, passt ein bisschen/manchmal zu mir, passt nicht zu mir)

86. Körperliches Erscheinen

		trifft zu	trifft etwas zu	trifft nicht zu
a	Ich bin schön.	☐(3)	☐(2)	☐(1)
b	Ich würde gerne anders aussehen.	☐(1)	☐(2)	☐(3)
c	Ich glaube, dass andere mich hässlich finden.	☐(1)	☐(2)	☐(3)
d	Ich bin sauber.	☐(3)	☐(2)	☐(1)
e	Ich ziehe mich gerne schön an.	☐(3)	☐(2)	☐(1)
f	Meine Grösse gefällt mir nicht.	☐(1)	☐(2)	☐(3)
g	Ich finde mich zu dick.	☐(1)	☐(2)	☐(3)
h	Ich bin zu dünn.	☐(1)	☐(2)	☐(3)

Fragen zur Geschlechtsidentität

Sehr gut, nun weiss ich mehr, was Du zu Deinem Körper findest. Nun würde ich gerne wissen, wie es für Dich ist, ein Junge zu sein.

87. Wie zufrieden bist Du, dass Du ein Junge und nicht ein Mädchen bist?

☐(3) sehr zufrieden ☐(2) zufrieden ☐(1) unzufrieden ☐(0) sehr unzufrieden

(Weiss nicht □(9))

88. Hast Du Dich schon mal mehr wie ein Mädchen als wie ein Junge gefühlt?

☐(1) ja ☐(0) nein (Weiss nicht □(9))

89. Hast Du schon einmal daran gedacht, dass Du kein richtiger Junge bist?

☐(1) ja ☐(0) nein (Weiss nicht □(9))

ab 12 J.: Fragen zur Sexualität

Ich möchte Dir jetzt einige Fragen zur Sexualität stellen. Einige Jugendliche haben bestimmte Erfahrungen schon gemacht und andere noch nicht. Bei diesen Fragen gibt es kein Richtig oder Falsch und vor allem kein Gut oder Schlecht!
Auch wenn es sich bei Sexualität um ein sehr persönliches Thema handelt, möchte ich Dich bitten, diese Fragen zu beantworten. Niemand erfährt, was Du antwortest. Die Antworten von Dir werde ich sehr vertraulich behandeln.

Zuerst möchte ich über das Verliebtsein sprechen

90. Hast Du Dich schon mal verliebt?

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein → ***weiter mit Frage 94***

(Weiss nicht □(9))

91. In wen hast Du Dich bisher verliebt?

☐(2) in Mädchen ☐(0) in Jungen ☐(1) in Mädchen und Jungen (Weiss nicht □(9))

92. Hast Du Dich schon mal mit einem Jungen/Mädchen verabredet, in den oder das Du verliebt warst?

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

93. Hattest Du schon einmal eine feste Freundin/einen festen Freund?

☐(1) ja, wieviele?_____ (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

Hast Du zurzeit eine feste Freundin? ☐(1) ja ☐(0) nein (Weiss nicht □(9))

Jetzt haben wir über das Verliebtsein gesprochen. Nun möchte ich etwas mehr über Deine sexuellen Erfahrungen im engeren Sinne wissen.

94. Hast Du als Kind Doktor gespielt? (*Wenn Kind Begriff nicht versteht, erklären, z. B. gegenseitig Körper und Geschlechtsorgane angucken)*

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

95. Hast Du Dich schon mal selbst befriedigt?

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

Befriedigst Du dich regelmässig selbst? ☐(1) ja ☐(0) nein (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

96. Bist Du bei der Selbstbefriedigung schon mal zum Orgasmus gekommen? (Ist bei der Selbstbefriedigung schon einmal Flüssigkeit aus Deinem Penis gekommen, was zu einem sehr befriedigenden Gefühl führte?)

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

97. Hast Du schon mal jemanden mit Zungenkuss geküsst?

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

98. Hast Du schon mal mit einem Mädchen/Jungen geschmust?

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

99. Hast Du schon mal Petting gemacht?

(Sich zärtlich streicheln, schmusen und sich auf diese Weise Lust machen, nennt man Petting. Man kann sich auch gegenseitig zum Orgasmus bringen und viel Spass haben)

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

100. Hattest Du schon mal Geschlechtsverkehr?

☐(1) ja → wie alt warst Du beim ersten Mal? ☐☐ Jahre (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

Nun möchte ich auch noch von Dir wissen, ob Du auch schon sexuelle Erfahrungen gemacht hast, die Du gar nicht wolltest.

101. Es gibt Menschen, von denen wird man gerne berührt und andere, von denen man nicht so gerne berührt wird. Hat Dich schon einmal jemand berührt, von dem Du es nicht wolltest?

☐(1) ja →Wer war das, der Dich berührt hat?_____ (Weiss nicht □(9))
Wo hat Dich diese Person überall berührt?_____ (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

102. Hast Du schon einmal jemanden berühren müssen, den Du nicht berühren wolltest?

☐(1) ja → Wen musstest Du berühren?_____ (Weiss nicht □(9))
Wo musstest Du diese Person berühren?_____ (Weiss nicht □(9))

☐(0) nein

(Weiss nicht □(9))

Zum Schluss stelle ich Dir noch eine Frage allgemein zur Sexualität. Bitte sage mir, ob die folgenden Aussagen auf Dich zutreffen. Du hast folgende Antwortmöglichkeiten zur Auswahl:…

103. Sexualität…

		trifft zu	**trifft etwas zu**	**trifft nicht zu**
a	…ist etwas Schönes.	☐(2)	☐(1)	☐(0)
b	…ist etwas Wichtiges.	☐(2)	☐(1)	☐(0)
c	…ist ein peinliches Thema.	☐(0)	☐(1)	☐(2)
d	…macht mir Angst.	☐(0)	☐(1)	☐(2)
e	…interessiert mich nicht.	☐(0)	☐(1)	☐(2)

Das waren nun alle Fragen, die ich Dir zum Thema Sexualität stellen wollte. Gibt es zur Sexualität noch etwas Wichtiges, das Du loswerden oder fragen möchtest?

bis 12 J.: Fragen zum Geschlechtsrollenverhalten

Im Folgenden stelle ich Dir Fragen zu Deinen Lieblingsbeschäftigungen und Vorlieben. Zu jeder Antwort gibt es drei Antworten. Du sollst diejenige auswählen, die am besten zu Dir passt.

104. Womit spielst Du am liebsten?

☐(3) Computer (Weiss nicht □(9))

☐(1) Verkleiden und Schminken

☐(2) Haustier

105. Was machst du am liebsten, wenn Du nicht in die Schule gehst?

☐(3) mit Lego spielen (Weiss nicht □(9))

☐(1) in der Küche kochen oder backen

☐(2) ein Buch oder einen Comic lesen

106. Was ist Dein Lieblingssport?

☐(3) Fussball (Weiss nicht □(9))

☐(2) Schwimmen

☐(1) Ballett

107. Was für Filme siehst Du am liebsten?

☐(3) spannende Filme (Weiss nicht □(9))

☐(2) Zeichentrickfilme

☐(1) romantische Filme

108. Was ist Deine Lieblingsrolle, wenn Ihr Familie spielt?

☐(3) Vater (Weiss nicht □(9))

☐(1) Mutter

☐(2) Kind

109. Als was gehst Du am liebsten an den Karneval, oder wenn Ihr Verkleiden spielt?

☐(1) Prinzessin (Weiss nicht □(9))

☐(3) Pirat

☐(2) Clown

110. Was möchtest Du später am liebsten werden?

☐(1) Krankenpfleger (Weiss nicht □(9))

☐(2) Reporter

☐(3) Polizist

111. Wenn du einen besten Freund oder eine beste Freundin auswählen musst, wählst Du dann:

☐(3) einen Jungen (Weiss nicht □(9))

☐(1) ein Mädchen

☐(2) ich weiss nicht, ob ich einen Jungen oder ein Mädchen wähle

112. Mit wem spielst Du in der Schule am liebsten?

☐(3) nur mit Jungen (Weiss nicht □(9))

☐(1) nur mit Mädchen

☐(2) mit Jungen und Mädchen

113. Was tust Du am liebsten?

☐(2) malen (Weiss nicht □(9))

☐(1) handarbeiten

☐(3) basteln

114. Macht es Dir Spass, neue Kleider zu kaufen?

☐(1) ja, immer (Weiss nicht □(9))

☐(2) manchmal

☐(3) nein, fast nie

115. Was ziehst Du am liebsten an?

☐(3) bequeme Sachen, die dreckig werden dürfen (Weiss nicht □(9))

☐(1) schöne, hübsche Sachen, die nicht schmutzig werden dürfen

☐(2) manchmal bequeme Sachen, manchmal hübsche Sachen

116. Wenn Du Geburtstag feierst, wen lädst Du dann am liebsten ein?

☐(3) nur Jungen (Weiss nicht □(9))

☐(1) nur Mädchen

☐(2) Jungen und Mädchen

117. Was machst Du am liebsten mit Deinen Freunden und Freundinnen?

☐(3) herumtoben (Weiss nicht □(9))

☐(2) Spiele spielen

☐(1) quatschen

118. Welchen Sport machst Du am liebsten?

☐(1) Eiskunstlaufen (Weiss nicht □(9))

☐(3) Judo

☐(2) Tennis

119. Wenn Du mit anderen Kindern was zusammen machen sollst, mit wem arbeitest Du dann am liebsten?

☐(3) nur mit Jungen (Weiss nicht □(9))

☐(1) nur mit Mädchen

☐(2) mit Jungen und Mädchen

120. Was machst Du meistens am liebsten?

☐(3) Fussball spielen (Weiss nicht □(9))

☐(1) einen Einkaufsbummel

☐(2) Fernsehen

121. Was möchtest Du später am liebsten werden?

☐(1) Friseur *(Coiffeur)* (Weiss nicht □(9))

☐(3) Tischler *(Schreiner)*

☐(2) Schauspieler

122. Was möchtest Du später am liebsten haben?

☐(3) ein Auto (Weiss nicht □(9))

☐(1) Kinder

☐(2) viel Geld

123. Wärst Du ab und zu gerne ein Mädchen?

☐(1) ja, sehr oft (Weiss nicht □(9))

☐(2) manchmal

☐(3) nein, nie

124. Wenn Du wütend bist, haust und trittst Du dann?

☐(3) ja, sehr oft (Weiss nicht □(9))

☐(2) manchmal

☐(1) nein, nie

125. Wenn Du traurig bist, weinst Du dann?

☐(1) ja, sehr oft (Weiss nicht □(9))

☐(2) manchmal

☐(3) nein, nie

126. Nun ***kommen 8 Sätze über Dich selbst. Sag mir wieder, ob der Satz zu Dir passt, manchmal passt oder nie passt.***

		ja, immer	**manchmal**	**nein, nie**
a	Ich bin stark.	☐(3)	☐(2)	☐(1)
b	Ich bin schüchtern.	☐(1)	☐(2)	☐(3)
c	Ich bin jungenhaft.	☐(3)	☐(2)	☐(1)
d	Ich bin hilfsbereit.	☐(1)	☐(2)	☐(3)
e	Ich bin der Boss.	☐(3)	☐(2)	☐(1)
f	Ich bin artig. *(brav).*	☐(1)	☐(2)	☐(3)
g	Ich bin mädchenhaft.	☐(1)	☐(2)	☐(3)
h	Ich bin trotzig.	☐(3)	☐(2)	☐(1)

127. ***Ich lese Dir nun noch eine kurze Geschichte vor. Anschliessend werde ich Dir zwei Fragen dazu stellen. Also***: Zwei Erfinder haben sehr lange an einem Auftrag gearbeitet. Sie wollten einen richtigen Menschen machen – also keinen Roboter, sondern einen echten Menschen aus Fleisch und Blut. Jetzt ist es endlich gelungen. Sie haben ein Kind von ungefähr 10 Jahren gemacht. Das Kind darf selbst wählen, ob es ein Junge oder ein Mädchen sein möchte. Das Kind findet die Entscheidung schwierig und bittet dich um Hilfe

a) Was würdest Du dem Kind sagen? □(3) werde ein Junge □(1)werde ein Mädchen

b) Erkläre Deine Antwort:

Interview-Beurteilung

Name der/des Interviewerin/Interviewers

Dauer des Interviews

Bemerkungen zum Auftreten und Verhalten des Kindes

Bemerkungen zum Auftreten und Verhalten der Eltern (Familie)

Qualität der Durchführung

☐(4) sehr gut ☐(3) gut ☐(2) mittel ☐(1) schlecht ☐(0) sehr schlecht

Allgemeine Bemerkungen zur Durchführung

Atmosphäre/Klima

Validitätseinschätzung von Antworten des Kindes:

☐(4) sehr gut ☐(3) gut ☐(2) mittel ☐(1) schlecht ☐(0) sehr schlecht

6.6 Appendix F: Mother's Questionnaire

Studie zum Langzeitverlauf bei Kindern und Jugendlichen nach Hypospadiekorrektur

ID-Nummer:_____

Fragebogen für die Mutter

Im Folgenden finden Sie eine Reihe von Fragen zur Hypospadie Ihres Sohnes, zu seinem Befinden und Verhalten, zu Ereignissen in der Familie, zu Ihrem Umgang mit Sexualität und zu Ihrem eigenen Befinden. Der Fragebogen beginnt mit einigen Fragen zu soziodemographischen Angaben zu Ihrer Person und Ihrer Familie.
Es ist wichtig, dass dieser Fragebogen von der Mutter des Kindes ausgefüllt wird. Bitte lesen Sie jede Frage sorgfältig durch. Machen Sie hinter jeder Frage ein Kreuz bei der für Sie am besten zutreffenden Antwort. Es kann sein, dass es Feststellungen gibt, bei denen keine der vorgesehenen Antworten genau auf Sie beziehungsweise auf Ihr Kind zutrifft. Bitte kreuzen Sie in diesem Fall diejenige Antwort an, die am ehesten zutrifft. Überlegen Sie bitte nicht erst, welche Antwort „den besten Eindruck" machen könnte, sondern antworten Sie möglichst spontan. Streichen Sie versehentliche Antworten deutlich durch und kreuzen Sie danach das richtige Kästchen an. Bitte beantworten Sie alle Fragen.

Wir versichern Ihnen, Ihre Angaben streng vertraulich zu behandeln.

Nachdem Sie alle Fragen beantwortet haben, bitten wir Sie, den Fragebogen der wissenschaftlichen Mitarbeiterin Frau Verena Schönbucher abzugeben oder mit dem beigelegten frankierten Couvert baldmöglichst (innerhalb 14 Tage) an uns zurückzusenden.

Herzlichen Dank!

Name und Vorname der Mutter:

Name und Vorname des Kindes:

Datum:

Angaben zu Ihrer Person und zu Ihrer Familie

Zunächst möchten wir Sie um einige Angaben zu Ihrer Person und zu Ihrer Familie bitten.

1. Sind Sie verheiratet oder leben Sie in einer festen Partnerschaft?

☐1 ja ☐0 nein

2. Wieviele Geschwister hat Ihr Kind? (gemeint sind auch Halb- und Adoptivgeschwister)

____ Bruder/Brüder

____ Schwester/n

3. Welche Staatsangehörigkeit haben Sie?

Mutter: ☐0 Schweiz

☐1 anderes Land, welches? _____

Vater: ☐0 Schweiz

☐1 anderes Land, welches? _____

Lebenspartner der Mutter / Stiefvater: ☐0 Schweiz

☐1 anderes Land, welches? _____

Lebenspartnerin des Vaters / Stiefmutter: ☐0 Schweiz

☐1 anderes Land, welches? _____

4. Welche Sprache wird bei Ihnen zu Hause meistens gesprochen?

☐0 deutsch

☐1 serbo-kroatisch

☐2 türkisch

☐3 italienisch

☐4 französisch

☐5 albanisch

☐6 andere, welche? _____

5. Gehören Sie einer Kirche / Glaubensgemeinschaft an?

☐0 nein

☐ ja, welcher? ☐1 römisch-katholische Kirche

☐2 reformierte / evangelische Kirche

☐3 christlich-orthodoxe Kirche

☐4 jüdische Glaubensgemeinschaft

☐5 islamische Glaubensgemeinschaft

☐6 andere, welche? _____

6. Welches ist der höchste Schulabschluss der Eltern des Kindes?

Mutter	**Vater**
☐1 Sonderschule/nicht abgeschlossene Mindestschulausbildung	☐1 Sonderschule/nicht abgeschlossene Mindestschulausbildung
☐2 Abschluss Mindestschulzeit	☐2 Abschluss Mindestschulzeit
☐3 Realschule	☐3 Realschule
☐4 Sekundarschule/Bezirksschule	☐4 Sekundarschule/Bezirksschule
☐5 Matura/Seminar/Fachhochschule	☐5 Matura/Seminar/Fachhochschule
☐6 Universität/ETH	☐6 Universität/ETH
☐7 anderer, bitte angeben:_____	☐7 anderer, bitte angeben:_____

7. Welches ist die höchste Berufsausbildung der Eltern des Kindes?

Mutter	**Vater**
☐1 ungelernt	☐1 ungelernt
☐2 angelernt	☐2 angelernt
☐3 2-jährige Lehre/Diplom/Abendschule	☐3 2-jährige Lehre/Diplom/Abendschule
☐4 3-4 jährige Lehre/Diplom/Handelsschule	☐4 3-4 jährige Lehre/Diplom/Handelsschule
☐5 Abschluss Seminar/Oberseminar/ Diplom Fachhochschule	☐5 Abschluss Seminar/Oberseminar/ Diplom Fachhochschule
☐6 Universität/ETH	☐6 Universität/ETH
☐7 andere, bitte angeben:_____	☐7 andere, bitte angeben:_____

8. Aktuelle berufliche Tätigkeiten der Eltern:

Mutter: _____

Vater: _____

9. Welche Schule besucht Ihr Kind zur Zeit?

☐1 Kindergarten

☐2 Regelschule (Primar-, Sekundar-, Real- oder Kantonsschule)

☐3 Berufsschule / Lehre

☐4 Kleinklasse

☐5 Sonderschule

☐6 Privatschule

☐7 Heilpädagogische Schule

☐8 andere, welche? _____

☐9 keine

10. Besuchte Ihr Kind ein zusätzliches Jahr den Kindergarten oder eine Einführungsklasse (1. Klasse in 2 Jahren)?

☐1 ja ☐0 nein

11. Hat Ihr Kind eine Klasse wiederholt?

☐1 ja ☐0 nein

12. Welche Schulnoten hatte ihr Kind im letzten Zeugnis in den folgenden Fächern?

Deutsch:_____

Mathematik:_____

Realien (Mensch und Umwelt):_____

Französisch (falls besucht):_____

13. Besucht Ihr Kind einen Stützunterricht oder erhält es andere Fördermassnahmen bezüglich seiner Schulleistung?

☐1 ja ☐0 nein

14. a) Wurde Ihr Kind jemals psychologisch/psychiatrisch untersucht oder behandelt?

☐1 ja, wann?_____welcher Grund?_____

☐0 nein

b) War Ihr Kind schon einmal wegen Unfalls oder Krankheit (ausser wegen seiner Hypospadie) im Spital hospitalisiert (mind. über 1 Nacht)?

☐1 ja, wann?_____welcher Grund?_____

☐0 nein

15. Besuchte Ihr Kind jemals eine der folgenden Therapien? Falls ja, welche und von wann bis wann (Jahr)?

a) Logopädie: ☐1 ja von:_____ bis:_____

☐0 nein

b) Psychomotorik: ☐1 ja von:_____ bis:_____

☐0 nein

c) Ergotherapie: ☐1 ja von:_____ bis:_____

☐0 nein

Fragen zur Hypospadie Ihres Kindes

Ihr Kind ist mit einer Hypospadie zur Welt gekommen, die am Kinderspital Zürich operativ korrigiert worden ist. In den anschliessenden Abschnitten möchten wir Sie gerne über die Hypospadie Ihres Kindes befragen.

16. Wann wurde bei Ihrem Kind die Diagnose Hypospadie gestellt?

☐0 unmittelbar nach der Geburt

☐1 später, wann?_____

17. Von wem wurde die Diagnose Hypospadie das erste Mal gestellt?

☐0 Geburtshelfer/in/Hebamme

☐1 behandelnde/r Kinderarzt/ärztin (Hausarzt/ärztin)

☐2 Kinderchirurg/in oder Urologe/in

☐3 von jemandem anderen, von wem? *(bitte keine Namen nennen)*_____

18. Wie alt war Ihr Kind zum Zeitpunkt als es von seiner Hypospadie bzw. seiner/n Operation/en erfuhr?

____Jahre

19. Aus welchem Anlass wurde Ihr Kind über seine Hypospadie informiert?

☐0 wir haben seine Hypospadie seit Geburt offen gelegt

☐1 als das Kind danach gefragt hat

☐2 als das Kind nach meinem Gefühl alt genug war

☐3 anderer Anlass, welcher?_____

20. Wie gut verstehen Sie gegenwärtig, was eine Hypospadie ist?

☐3 (sehr) gut ☐2 nicht so gut ☐1 ziemlich schlecht ☐0 schlecht

21. Haben Sie das Bedürfnis nach mehr Informationen zur Hypospadie Ihres Kindes?

☐1 ja ☐0 nein

22. Wir stark belastet Sie zurzeit die Hypospadie Ihres Kindes?

☐0 überhaupt nicht ☐1 ein wenig ☐2 ziemlich ☐3 stark ☐4 sehr stark

23. Machen Sie sich aufgrund der Hypospadie Ihres Sohnes Sorgen um folgende Dinge? *(Mehrfachnennungen möglich)*

	ja	**nein**
a) seine sexuelle Funktionsfähigkeit……..…....	☐1	☐0
b) seine Fertilität (Zeugungsfähigkeit)……..…	☐1	☐0
c) allfällige Schwierigkeiten auf der Partnersuche im Jugend- und Erwachsenenalter.....	☐1	☐0

24. Ist es schon vorgekommen, dass Sie mit der Nacktheit Ihres Sohnes anders umgegangen sind als mit derjenigen seiner Geschwister (z. B. dass Sie diese stärker vermieden haben)?

☐1 ja → Was haben Sie anders gemacht? _____

_____

☐0 nein

☐2 mein Sohn ist ein Einzelkind

25. War Ihnen die Hypospadie Ihres Sohnes jemals unangenehm oder peinlich?

☐1 ja → In welcher/n Situation/en? _____

_____

☐0 nein

26. Wie beurteilen Sie das erzielte Gesamtergebnis der Hypospadie-Operation/en Ihres Sohnes?

☐3 (sehr) gut ☐2 nicht so gut ☐1 ziemlich schlecht ☐0 schlecht

27. In der untenstehenden Tabelle sind verschiedene Merkmale des Penis Ihres Sohnes aufgelistet. Bitte kreuzen Sie bei jedem Merkmal an, wie zufrieden Sie damit sind.

	sehr zufrieden	**zufrieden**	**unzufrieden**	**sehr unzufrieden**
a) Penislänge…….	☐3	☐2	☐1	☐0
b) Position und Form der Harnröhrenöffnung..	☐3	☐2	☐1	☐0
c) Form der Eichel……	☐3	☐2	☐1	☐0
d) Form der Penishaut……..……..…	☐3	☐2	☐1	☐0
e) Achse des Penis (ob Erektion gerade ist)....	☐3	☐2	☐1	☐0
f) (Fehlen der) Vorhaut…….	☐3	☐2	☐1	☐0
g) Gesamteindruck des Penis……...…….	☐3	☐2	☐1	☐0

28. Wer hat bisher von der Hypospadie Ihres Sohnes erfahren? *(Mehrfachnennungen möglich)*

☐0 nur die Eltern

☐1 Geschwister des Kindes

☐2 Grosseltern des Kindes

☐3 andere Verwandte (z. B. Onkel, Tante)

☐4 Freunde des Kindes

☐5 Lehrer/in, Erzieher/in des Kindes

☐6 Schulklasse des Kindes

☐7 Freunde oder Bekannte der Eltern

☐8 andere Personen, wer? *(bitte keine Namen nennen)*_____

29. Gibt es Geschwister des Kindes oder andere Verwandte mit der Diagnose Hypospadie?

☐ ja, wer? ☐1 Vater

☐2 Grossvater väterlicherseits

☐3 Grossvater mütterlicherseits

☐4 Bruder

☐5 Onkel väterlicherseits

☐6 Onkel mütterlicherseits

☐7 Cousin väterlicherseits

☐8 Cousin mütterlicherseits

☐10 andere, wer? *(bitte keine Namen nennen)*_____

☐0 nein

☐2 weiss nicht

30. Ist ein anderer Knabe oder Mann aus Ihrer Familie beschnitten (Vorhaut entfernt)? *(Mehrfachnennungen möglich)*

☐ ja, wer? ☐1 Bruder/Halbbruder des Kindes

☐2 Vater/Stiefvater des Kindes

☐3 anderer Verwandter des Kindes (z. B. Onkel, Grossvater, Cousin)

☐0 nein

31. Gehören Sie einer Glaubensgemeinschaft oder einer Kultur an, in der Beschneidungen bei Jungen (Entfernung der Vorhaut) Tradition sind?

☐1 ja ☐0 nein

32. Für wie sinnvoll halten Sie Beschneidungen (Entfernung der Vorhaut) bei Jungen aus kulturellen bzw. religiösen Gründen?

☐4 sehr sinnvoll ☐3 sinnvoll ☐2 weder noch ☐1 sinnlos ☐0 sehr sinnlos

33. Für wie sinnvoll halten Sie Beschneidungen (Entfernung der Vorhaut) bei Jungen aus medizinischen Gründen?

☐4 sehr sinnvoll ☐3 sinnvoll ☐2 weder noch ☐1 sinnlos ☐0 sehr sinnlos

Gibt es in Bezug auf die Hypospadie Ihres Kindes noch Aspekte die Ihnen wichtig sind? Wenn ja, dann notieren Sie diese bitte hier:

_____

_____

_____

_____

Fragen zum Befinden Ihres Kindes

Auf den folgenden Seiten steht eine Anzahl von Fragen zum Befinden Ihres Kindes. Bei jeder Frage finden Sie ein paar Antworten. Wählen Sie bitte diejenige Antwort aus, die am besten zu Ihrem Kind passt. Setzen Sie dann ein Kreuzchen in das Kästchen dieser Antwort. Nachfolgend steht ein Beispiel.

Hatte Ihr Kind Kopfschmerzen?

nie ☒
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐	☐	☐	☐
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Wenn Ihr Kind in den vergangenen Wochen kein einziges Mal Kopfschmerzen hatte, setzen Sie ein Kreuzchen in das Kästchen vor "nie" wie im obigen Beispiel. Dann können Sie sofort mit der folgenden Frage weitermachen.
Wenn Ihr Kind manchmal oder oft Kopfschmerzen hatte, machen sie ein Kreuzchen bei einer dieser Antworten. Daneben steht die Frage "Mein Kind fühlte sich dabei". Tragen Sie dort ein, wie sich Ihr Kind fühlte, als es Kopfschmerzen hatte. Wie in dem nachfolgenden Beispiel, in welchem sich das Kind "nicht so gut" fühlte. Danach machen Sie mit der nächsten Frage weiter.

Hatte Ihr Kind Kopfschmerzen?

nie ☒
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐	☒	☐	☐
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Schmerzen und Beschwerden in den vergangenen Wochen
Denken Sie jeweils kurz darüber nach, wie es in den vergangenen Wochen war.

34. Hatte Ihr Kind gelegentlich Ohren- oder Halsschmerzen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

35. Hatte Ihr Kind gelegentlich Magen- oder Bauchschmerzen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

36. Hatte Ihr Kind gelegentlich Kopfschmerzen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

37. War Ihrem Kind gelegentlich schwindlig?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

38. War Ihrem Kind gelegentlich übel?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

39. War Ihr Kind müde?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

40. War Ihr Kind träge?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

41. War Ihr Kind benommen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

42. Hatte Ihr Kind andere Schmerzen oder Beschwerden?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Was für Schmerzen oder Beschwerden waren das?

Hatte Ihr Kind Schwierigkeiten mit den folgenden Aktivitäten in den letzen Wochen?
Denken Sie jeweils kurz darüber nach, wie es in den vergangenen Wochen war. Hatte Ihr Kind gelegentlich...

43. Mühe mit dem Gehen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

44. Mühe mit dem Laufen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

45. Mühe mit dem Stehen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

46. Mühe mit dem Heruntergehen von Treppen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

47. Mühe mit dem Spielen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

48. Mühe mit dem langen Gehen oder Laufen, mit der Ausdauer?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

49. Mühe mit dem Halten des Gleichgewichts?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

50. Mühe, Dinge geschickt und schnell zu erledigen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Hatte Ihr Kind Schwierigkeiten mit den folgenden Aktivitäten in den letzen Wochen?
Denken Sie jeweils kurz darüber nach, wie es in den vergangenen Wochen war. Hatte Ihr Kind gelegentlich...

51. Mühe, selbstständig zur Schule zu gehen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

52. Mühe, sich selbst zu waschen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

53. Mühe, sich selbst anzukleiden?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

54. Mühe, selbst zum WC zu gehen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

55. Mühe, selbst zu essen und zu trinken?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

56. Mühe, selbst Sport zu treiben oder draussen zu spielen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

57. Mühe, selbst Hobbys zu betreiben?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

58. Mühe mit dem Fahrradfahren?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Hatte Ihr Kind Schwierigkeiten mit den folgenden Aktivitäten in den letzen Wochen?
Denken Sie jeweils kurz darüber nach, wie es in den vergangenen Wochen war. Hatte Ihr Kind gelegentlich...

59. Mühe mit dem Aufpassen oder Konzentrieren?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

60. Mühe mit dem Begreifen der Schularbeit?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

61. Mühe mit dem Begreifen von dem, was andere gesagt haben?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

62. Mühe mit dem Rechnen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

63. Mühe mit dem Lesen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

64. Mühe mit dem Schreiben?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

65. Mühe mit dem Lernen?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

66. Mühe, die richtigen Worte zu finden?

nie ☐ (4)
manchmal ☐
oft ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

Mit andern Kindern und Ihnen selbst in den vergangenen Wochen
Denken Sie jeweils kurz darüber nach, wie es in den vergangenen Wochen war.

67. Mein Kind konnte mit anderen Kindern gut spielen oder sprechen.

ja ☐ (4)
nicht genug ☐
nie ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

68. Mein Kind konnte bei anderen Kindern sich selbst behaupten.

ja ☐ (4)
nicht genug ☐
nie ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

69. Andere Kinder baten mein Kind mitzuspielen.

ja ☐ (4)
nicht genug ☐
nie ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

70. Mein Kind fühlte sich bei anderen Kindern wohl.

ja ☐ (4)
nicht genug ☐
nie ☐

Mein Kind fühlte sich dabei:

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

71. Mein Kind konnte gut mit uns Eltern spielen oder sprechen.

ja	☐ (4)	
nicht genug	☐	Mein Kind fühlte sich dabei:
nie	☐	

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

72. Mein Kind war uns Eltern gegenüber schweigsam.

nie	☐ (4)	
manchmal	☐	Mein Kind fühlte sich dabei:
oft	☐	

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

73. Mein Kind war uns Eltern gegenüber unruhig oder ungeduldig.

nie	☐ (4)	
manchmal	☐	Mein Kind fühlte sich dabei:
oft	☐	

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

74. Mein Kind war uns Eltern gegenüber aufsässig.

nie	☐ (4)	
manchmal	☐	Mein Kind fühlte sich dabei:
oft	☐	

☐ (3)	☐ (2)	☐ (1)	☐ (0)
(sehr) gut	nicht so gut	ziemlich schlecht	schlecht

75. Mein Kind fühlte sich in den vergangenen Wochen…

	nie	**manchmal**	**oft**		**nie**	**manchmal**	**oft**
fröhlich:	☐0	☐1	☐2	entspannt:	☐0	☐1	☐2
betrübt:	☐2	☐1	☐0	aggressiv:	☐2	☐1	☐0
vergnügt:	☐0	☐1	☐2	glücklich:	☐0	☐1	☐2
böse:	☐2	☐1	☐0	wütend:	☐2	☐1	☐0
zufrieden:	☐0	☐1	☐2	selbstsicher:	☐0	☐1	☐2
besorgt:	☐2	☐1	☐0	eifersüchtig:	☐2	☐1	☐0
überschwenglich:	☐0	☐1	☐2	froh:	☐0	☐1	☐2
trübsinnig:	☐2	☐1	☐0	ängstlich:	☐2	☐1	☐0

Gibt es zum Befinden Ihres Kindes noch Aspekte, die Ihnen wichtig sind? Wenn ja, notieren Sie diese bitte hier:

Fragen zum Verhalten Ihres Kindes

*Auf den folgenden Seiten finden Sie eine Liste von Verhaltensweisen von Kindern/Jugendlichen. Bitte kreuzen Sie bei jeder Verhaltensweise diejenige Antwort an, die Ihr Kind **zur Zeit oder innerhalb der letzten sechs Monate** am besten beschreibt. Machen Sie bitte bei jedem Merkmal ein Kreuzchen. Falls Merkmale nicht zutreffen, kreuzen Sie das Kästchen "stimmt nicht" an.*

	Mein Kind:	**Stimmt nicht** (soweit Ihnen bekannt)	**Stimmt etwas** (od. manchmal)	**Stimmt genau** (od. häufig)
76.	Verhält sich zu jung für sein Alter…….....	☐0	☐1	☐2
77.	Hat Allergien…….	☐0	☐1	☐2
78.	Streitet oder widerspricht viel……...	☐0	☐1	☐2
79.	Hat Asthma……...	☐0	☐1	☐2
80.	Verhält sich wie ein Kind des anderen Geschlechts…….	☐0	☐1	☐2
81.	Kotet ein…….....	☐0	☐1	☐2
82.	Gibt an, schneidet auf…….....	☐0	☐1	☐2
83.	Kann sich nicht konzentrieren, kann nicht lange aufpassen	☐0	☐1	☐2

84.	Kommt von bestimmten Gedanken nicht los, hat Zwangsgedanken……..	☐0	☐1	☐2
85.	Kann nicht stillsitzen, ist unruhig oder überaktiv…….	☐0	☐1	☐2
86.	Klammert sich an Erwachsene oder ist abhängig…….	☐0	☐1	☐2
87.	Klagt über Einsamkeit…….....	☐0	☐1	☐2
88.	Ist verwirrt oder zerstreut…….…….	☐0	☐1	☐2
89.	Weint viel……..	☐0	☐1	☐2
90.	Ist roh zu den Tieren oder quält sie……...	☐0	☐1	☐2
91.	Ist roh oder gemein zu anderen, schüchtert sie ein……...	☐0	☐1	☐2
92.	Hat Tagträumereien oder ist gedankenverloren……	☐0	☐1	☐2
93.	Verletzt sich absichtlich oder versucht Selbstmord……..	☐0	☐1	☐2
94.	Verlangt viel Beachtung……	☐0	☐1	☐2
95.	Macht seine eigenen Sachen kaputt……..	☐0	☐1	☐2
96.	Macht Sachen kaputt, die den Eltern, Geschwistern oder anderen gehören……	☐0	☐1	☐2
97.	Gehorcht nicht zu Hause……...	☐0	☐1	☐2
98.	Gehorcht nicht in der Schule…….	☐0	☐1	☐2
99.	Isst schlecht……...	☐0	☐1	☐2
100.	Kommt mit anderen Kindern/Jugendlichen nicht gut aus….	☐0	☐1	☐2
101.	Scheint sich nicht schuldig zu fühlen, wenn es sich schlecht benommen hat…….	☐0	☐1	☐2
102.	Ist leicht eifersüchtig…….	☐0	☐1	☐2
103.	Isst oder trinkt Dinge, die nicht zum Essen oder Trinken geeignet sind (keine Süssigkeiten)……..…….	☐0	☐1	☐2
104.	Fürchtet sich vor bestimmten Tieren, Situationen oder Plätzen (Schule ausgenommen)……	☐0	☐1	☐2
105.	Hat Angst, in die Schule zu gehen……	☐0	☐1	☐2
106.	Hat Angst, etwas Schlimmes zu denken oder zu tun……	☐0	☐1	☐2
107.	Glaubt, perfekt sein zu müssen…….	☐0	☐1	☐2
108.	Beklagt sich, dass es nicht geliebt wird……	☐0	☐1	☐2
109.	Glaubt, andere wollen ihm etwas tun……	☐0	☐1	☐2
110.	Fühlt sich wertlos und unterlegen…….	☐0	☐1	☐2
111.	Verletzt sich häufig und ungewollt, neigt zu Unfällen…….	☐0	☐1	☐2
112.	Gerät leicht in Raufereien, Schlägereien……...	☐0	☐1	☐2
113.	Wird viel gehänselt……...	☐0	☐1	☐2

		Stimmt nicht (soweit Ihnen bekannt)	**Stimmt etwas** (od. manchmal)	**Stimmt genau** (od. häufig)
114.	Hat Umgang mit anderen, die in Schwierigkeiten geraten...	☐0	☐1	☐2
115.	Hört Geräusche und Stimmen, die nicht da sind……...	☐0	☐1	☐2
116.	Ist immer impulsiv oder handelt, ohne zu überlegen……	☐0	☐1	☐2
117.	Ist lieber allein als mit anderen zusammen……...…	☐0	☐1	☐2
118.	Lügt, betrügt oder schwindelt……...	☐0	☐1	☐2
119.	Kaut Fingernägel……...	☐0	☐1	☐2
120.	Ist nervös oder angespannt……	☐0	☐1	☐2
121.	Hat nervöse Bewegungen oder Zuckungen……..	☐0	☐1	☐2
122.	Hat Albträume……...	☐0	☐1	☐2
123.	Ist bei anderen Kindern oder Jugendlichen nicht beliebt…..	☐0	☐1	☐2
124.	Leidet an Verstopfung……...	☐0	☐1	☐2
125.	Ist zu furchtsam oder ängstlich……	☐0	☐1	☐2
126.	Fühlt sich schwindelig……...……...	☐0	☐1	☐2
127.	Hat zu starke Schuldgefühle……...……..	☐0	☐1	☐2
128.	Isst zu viel…….	☐0	☐1	☐2
129.	Ist immer müde…….	☐0	☐1	☐2
130.	Hat Übergewicht……...	☐0	☐1	☐2
131.	Hat folgende Beschwerden ohne bekannte körperliche Ursache:			
a)	Schmerzen (ausser Kopf- und Bauchschmerzen)…….…	☐0	☐1	☐2
b)	Kopfschmerzen…….	☐0	☐1	☐2
c)	Übelkeit…….	☐0	☐1	☐2
d)	Augenbeschwerden (ausgenommen solche, die durch Brille korrigiert sind)……	☐0	☐1	☐2
e)	Hautausschläge oder andere Hautprobleme……..	☐0	☐1	☐2
f)	Bauchschmerzen oder Magenkrämpfe……..	☐0	☐1	☐2
g)	Erbrechen……..	☐0	☐1	☐2
h)	Andere Beschwerden Bitte beschreiben (und ankreuzen):……...	☐0	☐1	☐2
	--- ---			
132.	Greift andere körperlich an……...	☐0	☐1	☐2

	Stimmt nicht (soweit Ihnen bekannt)	**Stimmt etwas** (od. manchmal)	**Stimmt genau** (od. häufig)
133. Bohrt in der Nase, zupft oder kratzt sich an anderen Körperstellen…….	☐0	☐1	☐2
134. Spielt in der Öffentlichkeit an den eigenen Geschlechtsteilen……..	☐0	☐1	☐2
135. Spielt zu viel an den eigenen Geschlechtsteilen……...	☐0	☐1	☐2
136. Ist schlecht in der Schule…….....	☐0	☐1	☐2
137. Ist körperlich unbeholfen oder ungeschickt……..	☐0	☐1	☐2
138. Ist lieber mit älteren Kindern oder Jugendlichen als mit Gleichaltrigen zusammen…….	☐0	☐1	☐2
139. Ist lieber mit jüngeren Kindern zusammen…….....	☐0	☐1	☐2
140. Weigert sich zu sprechen…….....	☐0	☐1	☐2
141. Tut bestimmte Dinge immer und immer wieder, wie unter einem Zwang…….	☐0	☐1	☐2
142. Läuft von zu Hause weg……	☐0	☐1	☐2
143. Schreit viel……	☐0	☐1	☐2
144. Ist verschlossen, behält Dinge für sich…….	☐0	☐1	☐2
145. Sieht Dinge, die nicht da sind……...	☐0	☐1	☐2
146. Ist befangen oder wird leicht verlegen……..	☐0	☐1	☐2
147. Zündet gerne oder hat schon Feuer gelegt……..….	☐0	☐1	☐2
148. Hat sexuelle Probleme…….....	☐0	☐1	☐2
149. Produziert sich gerne oder kaspert rum…….	☐0	☐1	☐2
150. Ist schüchtern oder zaghaft……..……	☐0	☐1	☐2
151. Schläft weniger als die meisten Gleichaltrigen…….	☐0	☐1	☐2
152. Schläft tagsüber und/oder nachts mehr als die meisten Gleichaltrigen……	☐0	☐1	☐2
153. Schmiert oder spielt mit Kot……..……...	☐0	☐1	☐2
154. Hat Schwierigkeiten beim Sprechen…….	☐0	☐1	☐2
155. Starrt ins Leere……..	☐0	☐1	☐2
156. Stiehlt zu Hause……...…….	☐0	☐1	☐2
157. Stiehlt anderswo……	☐0	☐1	☐2
158. Hortet Dinge, die es gar nicht braucht……..……...	☐0	☐1	☐2
159. Verhält sich seltsam oder eigenartig……..……..	☐0	☐1	☐2

	Stimmt nicht (soweit Ihnen bekannt)	**Stimmt etwas** (od. manchmal)	**Stimmt genau** (od. häufig)
160. Hat seltsame Gedanken oder fremdartige Ideen und Vorstellungen……	☐0	☐1	☐2
161. Ist störrisch, mürrisch oder reizbar……...…	☐0	☐1	☐2
162. Zeigt plötzliche Stimmungs- und Gefühlswechsel……...…	☐0	☐1	☐2
163. Schmollt viel und ist leicht eingeschnappt……	☐0	☐1	☐2
164. Ist misstrauisch……..	☐0	☐1	☐2
165. Flucht oder gebraucht obszöne (schmutzige) Wörter……...	☐0	☐1	☐2
166. Spricht davon, sich umzubringen……..	☐0	☐1	☐2
167. Redet oder wandelt im Schlaf……..……	☐0	☐1	☐2
168. Redet zu viel……..	☐0	☐1	☐2
169. Hänselt andere gern……...	☐0	☐1	☐2
170. Hat Wutausbrüche oder ein hitziges Temperament…...…...	☐0	☐1	☐2
171. Denkt zu viel an Sex……...……..	☐0	☐1	☐2
172. Bedroht andere……...……...	☐0	☐1	☐2
173. Lutscht am Daumen……...……...	☐0	☐1	☐2
174. Ist sehr auf Ordentlichkeit und Sauberkeit bedacht……..	☐0	☐1	☐2
175. Hat Schwierigkeiten mit dem Schlafen…….	☐0	☐1	☐2
176. Schwänzt die Schule (auch einzelne Stunden)……..	☐0	☐1	☐2
177. Zeigt zu wenig Aktivität, ist zu langsam oder träge…….	☐0	☐1	☐2
178. Ist unglücklich, traurig oder niedergeschlagen…….....	☐0	☐1	☐2
179. Ist ungewöhnlich laut……	☐0	☐1	☐2
180. Trinkt Alkohol, nimmt Drogen oder missbraucht Medikamente……	☐0	☐1	☐2
181. Richtet mutwillig Zerstörung an……...…	☐0	☐1	☐2
182. Nässt tagsüber ein……..…..	☐0	☐1	☐2
183. Nässt im Schlafen ein……	☐0	☐1	☐2
184. Quengelt oder jammert……..	☐0	☐1	☐2
185. Möchte gern ein Kind/Jugendlicher des anderen Geschlechts sein……	☐0	☐1	☐2
186. Zieht sich zurück, nimmt keinen Kontakt mit anderen auf...	☐0	☐1	☐2
187. Macht sich zu viele Sorgen……..……	☐0	☐1	☐2

		Stimmt nicht (soweit Ihnen bekannt)	Stimmt etwas (od. manchmal)	Stimmt genau (od. häufig)
188.	Bitte beschreiben sie hier die Probleme Ihres Kindes, die bisher noch nicht erwähnt wurden (und ankreuzen):……..…… ----------	☐0	☐1	☐2

Gibt es zum Verhalten Ihres Kindes noch Aspekte, die Ihnen wichtig sind? Wenn ja, notieren Sie diese bitte hier:

Fragen zur Sexualität

Im Folgenden möchten wir Sie zur sexuellen Entwicklung Ihres Kindes und zu Ihrem Umgang mit Sexualität in Ihrer Familie befragen. Bitte kreuzen Sie bei den nachfolgenden Aussagen an, wie sehr diese auf Sie/Ihre Familie/Ihr Kind zutreffen. Überlegen Sie bitte nicht erst, welche Antwort „den besten Eindruck" machen könnte, sondern antworten Sie so, wie es für Sie/ Ihre Familie/Ihr Kind persönlich zutrifft.

		stimmt nicht	stimmt etwas	stimmt genau
189.	Mit meinem/r Partner/in tausche ich täglich Zärtlichkeiten aus…	☐0	☐1	☐2
190.	Mit meinem Partner tausche ich nur Zärtlichkeiten aus, wenn die Kinder nicht anwesend sind……..	☐2	☐1	☐0
191.	Mein Kind ist in sexuellen Dingen aufgeklärt……..	☐0	☐1	☐2
192.	Mein Kind hat schon öfters von sich aus von sexuellen Dingen gesprochen…….	☐0	☐1	☐2
193.	Ich habe schon öfters mit meinem Kind über Sexualität gesprochen…….	☐0	☐1	☐2
194.	Bevor Kinder in die Pubertät kommen, können sie Informationen über Sexualität nicht verstehen……..	☐2	☐1	☐0
195.	Es ist (auch) Aufgabe der Eltern, die eigenen Kinder aufzuklären……	☐0	☐1	☐2
196.	Selbstbefriedigung von Kindern ist unnatürlich und ungesund….	☐2	☐1	☐0
197.	Kinder im Vorschulalter sollten sich nicht für sexuelle Dinge interessieren……..	☐2	☐1	☐0
198.	Ich finde es in Ordnung, wenn Kleinkinder beim Wickeln oder Umziehen mit ihren eigenen Genitalien spielen……	☐0	☐1	☐2
199.	Es ist wichtig, dass man mit dem eigenen Kind von klein auf offen über Sexualität spricht……..	☐0	☐1	☐2

	stimmt nicht	stimmt etwas	stimmt genau
200. Kinder sollten von ihren Eltern ermutigt werden, Fragen zur Sexualität zu stellen……...	☐0	☐1	☐2
201. Aufklärung bedeutet, dass Kind darüber zu informieren, wie ein Baby gezeugt wird…….	☐2	☐1	☐0
202. Sexuelle Handlungen von Kindern darf man nicht bestrafen..…..	☐0	☐1	☐2
203. Ich toleriere Menschen mit anderen sexuellen Lebensstilen, solange dabei niemandem zu Schaden kommt……..	☐0	☐1	☐2

204. Hat Ihr Kind Ihren Wissens jemals negative sexuelle Erlebnisse gemacht?

☐1 ja, welche? _____

_____

☐0 nein

Gibt es in Bezug auf die sexuelle Entwicklung Ihres Kindes oder bezüglich Ihrer Einstellung zur Sexualität noch Aspekte die Ihnen wichtig sind? Wenn ja, dann notieren Sie diese bitte hier:

_____

_____

_____

_____

_____

Fragen zum Familienleben

Nun möchten wir auch noch etwas über Ihr Familienleben erfahren. Sie finden nachfolgend verschiedene Aussagen über das Familienleben. Wenn eine Aussage Ihre Familie treffend beschreibt, kreuzen Sie bitte "stimmt" an, wenn die Aussage nicht auf Ihre Familie zutrifft, machen Sie ein Kreuzchen in das Kästchen "stimmt nicht". Bei Unentschiedenheit kreuzen Sie die Aussage "stimmt etwas" an.

	stimmt nicht	stimmt etwas	stimmt
205. Familienmitglieder helfen und unterstützen einander wirklich……...	☐0	☐1	☐2
206. Familienmitglieder behalten ihre Gefühle oft für sich…….....	☐0	☐1	☐2
207. Wir streiten uns häufig in unserer Familie……..……	☐0	☐1	☐2
208. Es scheint, als ob wir zu Hause oft die Zeit totschlagen……..……...	☐0	☐1	☐2
209. Wir sagen zu Hause alles, was wir wollen……..……	☐0	☐1	☐2
210. Familienmitglieder werden selten offen ärgerlich……..…….	☐0	☐1	☐2
211. Wir investieren eine Menge Energie in das, was wir zu Hause tun.....	☐0	☐1	☐2
212. Es ist schwierig, zu Hause Dampf abzulassen, ohne jemanden aufzuregen……..….	☐0	☐1	☐2

	stimmt nicht	stimmt etwas	stimmt
213. Familienmitglieder werden manchmal so wütend, dass sie Dinge herumwerfen…….…..	☐0	☐1	☐2
214. In unserer Familie gibt es ein Gefühl von Zusammengehörigkeit….....	☐0	☐1	☐2
215. Wir erzählen einander unsere persönlichen Probleme……..……..	☐0	☐1	☐2
216. Familienmitglieder verlieren kaum einmal ihre Fassung…….....	☐0	☐1	☐2
217. Wir melden uns selten freiwillig, wenn zu Hause etwas getan werden muss……..……...	☐0	☐1	☐2
218. Wenn wir Lust haben, etwas aus dem Augenblick heraus zu machen, machen wir uns oft einfach auf und gehen……..……	☐0	☐1	☐2
219. Familienmitglieder kritisieren einander häufig……..…….....	☐0	☐1	☐2
220. Familienmitglieder stärken einander wirklich den Rücken……..…...	☐0	☐1	☐2
221. Jemand in unserer Familie regt sich gewöhnlich auf, wenn Du Dich beklagst……..…..	☐0	☐1	☐2
222. Familienmitglieder schlagen einander manchmal……..…..…..	☐0	☐1	☐2
223. Es gibt sehr wenig Gruppengeist in unserer Familie……..…..…..	☐0	☐1	☐2
224. In unserer Familie sprechen wir offen über Geld und das Bezahlen von Rechnungen…….……	☐0	☐1	☐2
225. Wenn es eine Meinungsverschiedenheit gibt in unserer Familie, versuchen wir angestrengt, die Dinge auszuglätten und den Frieden zu bewahren……..……...	☐0	☐1	☐2
226. Wir kommen wirklich gut miteinander aus……..……..……	☐0	☐1	☐2
227. Wir sind üblicherweise vorsichtig in Bezug auf das, was wir einander sagen……..……..	☐0	☐1	☐2
228. Familienmitglieder versuchen einander oft voraus zu sein oder einander zu übertreffen……..……..	☐0	☐1	☐2
229. Es gibt in unserer Familie reichlich Zeit und Aufmerksamkeit für jeden Einzelnen……...…….	☐0	☐1	☐2
230. Es gibt eine Menge spontaner Diskussionen in unserer Familie..…...…	☐0	☐1	☐2
231. Wir glauben, dass man in unserer Familie mit einer lauten Stimme nichts erreicht……..…	☐0	☐1	☐2

Gibt es in Bezug auf Ihr Familienleben noch Aspekte die Ihnen wichtig sind? Wenn ja, dann notieren Sie diese bitte hier:

Fragen zu bedeutsamen Lebensereignissen

*In der Folge möchten wir wissen, welche bedeutsamen Lebensereignisse **Sie** erlebt haben. Bitte geben Sie in der nachfolgenden Liste an, ob in den **letzten 12 Monaten** eines der folgenden Ereignisse in Ihrer Familie stattgefunden hat.*

	nein	**ja**
232. Eigene Schwangerschaft oder Geburt…….	☐0	☐1
233. Eigene Trennung oder Scheidung……...	☐0	☐1
234. Eigene Heirat / Wiederverheiratung…….....	☐0	☐1
235. Zuzug einer verwandten oder bekannten Person in den eigenen Haushalt……..	☐0	☐1
236. Deutliche Veränderung des Familieneinkommens……..…	☐0	☐1
237. Ernsthafte Verschuldung……..	☐0	☐1
238. Umzug / Wohnortswechsel……..…	☐0	☐1
239. Eigener Stellenwechsel oder Stellenwechsel des Ehepartners……..…..	☐0	☐1
240. Eigene Arbeitslosigkeit oder Arbeitslosigkeit des Ehepartners……..…….	☐0	☐1
241. Ernsthafte Krankheit oder schwerer Unfall eines Familienmitgliedes (ausser Ihres Kindes)……...	☐0	☐1
242. Todesfall in der Familie oder im engeren Bekanntenkreis……..……	☐0	☐1
243. Schulwechsel eines Ihrer Kinder……..…...	☐0	☐1

Gibt es in Bezug auf bedeutende Ereignisse in Ihrem Leben noch Aspekte die Ihnen wichtig sind? Wenn ja, dann notieren Sie diese bitte hier:

Fragen zum Befinden der Mutter

*Folgende Fragen sollen ausschliesslich **von der Mutter** des Kindes **ausgefüllt werden!***
*Sie finden im Folgenden eine Liste von Problemen und Beschwerden, die man manchmal hat. Bitte lesen Sie jede Frage sorgfältig durch und entscheiden Sie, wie sehr Sie **in den letzten sieben Tagen** durch diese Beschwerden gestört oder bedrängt worden sind. Überlegen Sie bitte nicht erst, welche Antwort "den besten Eindruck" machen könnte, sondern antworten Sie so, wie es für Sie persönlich zutrifft. Machen Sie bitte hinter jeder Frage nur ein Kreuz in das Kästchen mit der für Sie am besten zutreffenden Antwort. **Bitte beantworten Sie jede Frage.***

Wie sehr litten Sie in den letzten sieben Tagen unter...?

	überhaupt nicht	ein wenig	ziemlich	stark	sehr stark
244. starker Befangenheit im Umgang mit anderen……..…	☐0	☐1	☐2	☐3	☐4
245. Schwermut…….....	☐0	☐1	☐2	☐3	☐4
246. Befürchtungen, wenn Sie alleine aus dem Haus gehen…..	☐0	☐1	☐2	☐3	☐4
247. Furchtsamkeit……..	☐0	☐1	☐2	☐3	☐4
248. Gedanken an den Tod und ans Sterben……..……	☐0	☐1	☐2	☐3	☐4
249. Leere im Kopf……...……..	☐0	☐1	☐2	☐3	☐4
250. Gedächtnisschwierigkeiten……...……..	☐0	☐1	☐2	☐3	☐4
251. dem Gefühl, dass die Leute unfreundlich sind oder Sie nicht leiden können…….	☐0	☐1	☐2	☐3	☐4
252. Energielosigkeit oder Verlangsamung in den Bewegungen oder im Denken……..…..	☐0	☐1	☐2	☐3	☐4
253. Übelkeit oder Magenverstimmung……...…..	☐0	☐1	☐2	☐3	☐4
254. Hitzewallungen oder Kälteschauern……...…	☐0	☐1	☐2	☐3	☐4
255. mangelnder Anerkennung Ihrer Leistungen durch andere……..	☐0	☐1	☐2	☐3	☐4
256. Ohnmachts- oder Schwindelgefühlen……...……..	☐0	☐1	☐2	☐3	☐4
257. dem Gefühl, dass die Leute Sie ausnutzten, wenn Sie es zulassen würden.....	☐0	☐1	☐2	☐3	☐4
258. einem Gefühl der Hoffnungslosigkeit angesichts der Zukunft……	☐0	☐1	☐2	☐3	☐4
259. dem Gefühl, einen Klumpen (Kloss) im Hals zu haben….	☐0	☐1	☐2	☐3	☐4
260. dem Gefühl, dass man den meisten Menschen nicht trauen kann……..	☐0	☐1	☐2	☐3	☐4
261. Herzklopfen oder Herzjagen……...…	☐0	☐1	☐2	☐3	☐4

		überhaupt nicht	ein wenig	ziemlich	stark	sehr stark
262.	Ideen oder Anschauungen, die andere nicht mit Ihnen teilen……	☐0	☐1	☐2	☐3	☐4
263.	der Furcht, in der Öffentlichkeit in Ohnmacht zu fallen….	☐0	☐1	☐2	☐3	☐4
264.	Minderwertigkeitsgefühlen gegenüber anderen……..	☐0	☐1	☐2	☐3	☐4
265.	Gedanken, sich das Leben zu nehmen……...…….	☐0	☐1	☐2	☐3	☐4
266.	einem unbehaglichen Gefühl, wenn Leute Sie beobachten oder über Sie reden……..…...	☐0	☐1	☐2	☐3	☐4
267.	Konzentrationsschwierigkeiten……..	☐0	☐1	☐2	☐3	☐4
268.	der Notwendigkeit, bestimmte Dinge, Orte oder Tätigkeiten zu meiden, weil Sie durch diese erschreckt werden….	☐0	☐1	☐2	☐3	☐4
269.	Schwierigkeiten beim Atmen……..	☐0	☐1	☐2	☐3	☐4
270.	Furcht auf offenen Plätzen oder auf der Strasse……..…..	☐0	☐1	☐2	☐3	☐4

Gibt es in Bezug auf Ihr Befinden noch Aspekte die Ihnen wichtig sind? Wenn ja, dann notieren Sie diese bitte hier:

Herzlichen Dank für Ihre Mitarbeit!

Sie haben uns mit Ihren Angaben sehr geholfen und einen wichtigen Beitrag für die zukünftige Behandlung und Betreuung von Kindern und Jugendlichen mit einer Hypospadie geleistet!

Falls Sie über die Ergebnisse dieser Studie informiert werden wollen, dann kreuzen Sie bitte ein letztes Mal mit ja an:

□ ja

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Curriculum Vitae

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EDUCATION

2008	Doctorate in Psychology (PhD), University of Zurich, Switzerland Doctoral thesis: Psychosexual development, psychosocial adjustment and quality of life of children and adolescents with hypospadias
2005	Licentiate (equivalent to Masters of Science) in Psychology, Psychopathology (adulthood), Social and Economic History, University of Zurich, Switzerland Master thesis (transl.): Lack of sexual desire of women from the age of 45
1998	Federal Maturity (Typus B), Kantonsschule am Burggraben, St. Gallen, Switzerland

WORK EXPERIENCE

Postgraduate

2007 – present	Research Fellow at the University Hospital Hamburg-Eppendorf, Department of Sex Research and Forensic Psychiatry, Germany Scholarship holder of the Swiss National Science Foundation (SNSF)

Conducting research on sexual quality of life of patients with disorders of sex development (DSD) in the Hamburg Intersex Project under supervision of Prof. Hertha Richter-Appelt, supervising master theses, co-organizing of the national congress "Interdisziplinäres Forum zur Intersexualität", 30th January 2008, University Hospital Hamburg-Eppendorf

2005 – 2007 Research Fellow at the University Children's Hospital Zurich, Pediatric Urology, Switzerland

Conducting a study on the psychosexual development, psychosocial adjustment and quality of life of children and adolescents with hypospadias under supervision of Daniel Weber, MD and Markus Landolt, Ph.D (Funding source: Foundation Mercator Switzerland)

2005 Social pedagogue and psychologist in the Children's Home Effingen (AG), Switzerland

Teaching and parenting children and adolescents

As a student

2005 Research Assistant at the Department of Psychology, Applied Cognitive Psychology, University of Zurich, Switzerland (Prof. Dr. D. Läge)

Compiling literature for the project «Psychopathology Taught Online»

2001-2004 Research and Teaching Assistant at the Department of Psychology, Social- and Health Psychology, University of Zurich, Switzerland (Prof. Dr. R. Hornung)

Tutoring undergraduate students, assistance in several surveys, compiling literature for seminaries

2002-2004 Private tutor in statistics

Training undergraduate students for statistics exams

2001-2002 Several practical trainings at the following institutions:
Psychiatric University Hospital Zurich, Switzerland
Psychiatric Hospital Meilen (ZH), Switzerland
Institute for social therapy Egliswil (AG), Switzerland
Day nursery Colibri, Zurich, Switzerland

Conducting psychiatric and psychodiagnostic interviews, conducting psychometric tests, psychological caring, supporting and partly nursing of patients, writing of psychological reports, conducting work therapy, co-organizing and conducting of excursions and camps, conducting night services, child caring

1999–2005 Assistant in a private psychotherapeutic joint practice

Psychological caring and psychosocial supporting of clients, writing of psychological reports, participation in individual psychotherapy sessions, in group psychotherapy sessions, in workshops and supervision sessions

AWARDS AND SCHOOLARSHIPS

2007-2008 Scholarship of the Swiss National Science Foundation (SNSF).

LANGUAGES

German: Native language

English: Proficiency level

French: Advanced knowledge

Spanish: Preliminary knowledge

PUBLICATIONS AND PRESENTATIONS

Publications

Schönbucher, V. (2007). Sexuelle Zufriedenheit von Frauen: Psychosoziale Faktoren. Eine Übersichtsarbeit. *Zeitschrift für Sexualforschung*, *1*, 21-41.

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Schweizer, K., Brinkmann, L., **Schönbucher, V.**, & Richter-Appelt, H. (im Druck). *Behandlungserfahrungen, Partnerschaft und Sexualität bei XY-Frauen – Ergebnisse der Hamburger Studie zur Intersexualität.* Jahresband der Deutschen Gesellschaft für Psychosomatische Frauenheilkunde und Geburtshilfe (DGPFG)

Richter-Appelt, H., **Schönbucher, V.**, & Schweizer, K. (im Druck). Betreuung erwachsener Patientinnen mit Intersexualität. *Gynäkologie und Geburtshilfe*

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Richter-Appelt, H., **Schönbucher, V.**, & Schweizer, K. (im Druck). Transsexualität versus Intersexualität: zwei unterschiedliche Varianten der Geschlechtsentwicklung. *Gynäkologie und Geburtshilfe*

Presentations

Richter-Appelt, H., Schweizer, K., & **Schönbucher, V.** (2008). Behandlungserfahrungen, psychisches Befinden und körperliches Wohlbefinden bei erwachsenen Personen mit Intersexualität. Vortrag am Interdisziplinären Forum zur Intersexualität (DSD), Hamburg, 30. Januar 2008

Schönbucher, V. B., Landolt, M. A., Gobet, R., & Weber, D. (2007). Psychosexual development of children and adolescents with hypospadias. Oral presentation at the ISHID 2nd

World Congress on Hypospadias and Disorders of Sex Development (DSD), Rome, 16-18th Nov 2007.

Schönbucher, V. B., Weber, D. M., Gobet, R., & Landolt, M. A. (2007). Psychosexuelle Entwicklung, psychisches Befinden und Lebensqualität von Kindern und Jugendlichen mit einer Hypospadie. Gastvortrag am Institut für Sexualforschung und Forensische Psychiatrie, 7. Oktober 2007

Weber, D. M., **Schönbucher, V. B.**, Gobet, R., & Landolt, M. A. (2007). Self-perception of genitalia after hypospadia repair. Vortrag an der 37. Jahresversammlung der Schweizerischen Gesellschaft für Kinderchirurgie, Bern, 4. Oktober 2007.

Schönbucher, V. B., Landolt, M. A., Gobet, R., & Weber, D. (2007). Health-related quality of life and psychological adjustment of children and adolescents with hypospadias. Oral presentation at the 20th International Symposium on Pediatric Research, Berne, 5-6th Oct 2007.

Schönbucher, V. B., Weber, D. M., & Landolt, M. A. (2007). Die gesundheitsbezogene Lebensqualität von Kindern und Jugendlichen mit einer Hypospadie. Vortrag am 8. Kongress für Gesundheitspsychologie, Schwäbisch Gmünd, 17.-19. September 2007.

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Schönbucher, V.B., Weber, D.M., & Landolt, M.A. (2007). Psychosoziale Aspekte der Hypospadie. Vortrag an der 58. Jahrestagung des Deutschen Kollegiums für Psychosomatische Medizin/15. Jahrestagung der Deutschen Gesellschaft für Psychosomatische Medizin und Ärztliche Psychotherapie, Nürnberg, 21.-24. März 2007.

Schönbucher, V.B., Weber, D.M., & Landolt, M.A. (2006). Quality of life of children and adolescents with hypospadias: Preliminary results. Poster presentation at the congress „From Gene to Gender. 2nd International Symposium on Disorders of Sex Development“, Lubeck, 30 th Aug - 2 th Sept 2006

Schönbucher, V. (2005). Sexuelle Lustlosigkeit von Frauen ab 45 Jahren. Vortrag an der Tagung «Work in Progress, Gender Studies», Zürich, 25. Mai 2005

Schönbucher, V. & Bucher, T. (2004). Sexuelle Lustlosigkeit von Frauen ab 45 Jahren. Posterpräsentation am Lizentianden- und Doktoranden-Kongress (LIDOKO) des Psychologischen Instituts der Universität Zürich, 24. Juni 2004